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INTEGRATED RADIOLOGY*

PRESIDENT'S ADDRESS

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IT IS with a sense of great humility that at this, the Sixty-second Annual Meeting, I assume the Presidency of the American Roentgen Ray Society. To be elected President of the oldest radiologic organization in America is a coveted distinction far beyond my desert and I should like first of all to express my profound gratitude for the honor bestowed upon me. My ardent hope is that I may prove worthy of the confidence placed in me and that I may be able to serve you conscientiously in the tradition established by a long array of illustrious predecessors and so laudably exemplified by Dr. Reineke, my immediate predecessor.

Dr. Reineke, in his scholarly dissertation of last year,[1] called attention to certain "trends and attitudes" in radiologic practice, which have developed during recent years and he admonished all of us not to sail our bark of ambition into the "trade winds" which may lead us onto the shoals of illusion and into the banks of disappointment, but rather we should steer into the quiet coves of refuge where we might meditate and consider the present and future direction of our voyage.

While Dr. Reineke spoke on the important aspect of the over-all practice of radiology and the ethics involved, of equal importance are the trends and attitudes in regard to future organizational set-ups of radiology in our country. With this view in mind I have chosen as a topic of my Inaugural Address "Integrated Radiology." I have chosen this topic for two reasons. First, the discovery of man-made radiations in 1895 by Röntgen, soon followed by the discovery of earthborn natural radiations and later by the development of artificially produced radiations, has resulted in the short span of 65 years in such amazingly rapid progress and vast expansion of the medical utilization of these radiations that, although Radiology was recognized early as a Specialty, certain trends were noted and attitudes expounded to divide this Specialty into three major disciplines, independent from each other, and more recently some consideration is being given to even further division into various subspecialties. Secondly, our Society, historically, reflects better than any other radio-

[1] REINEKE, H. G. Whither bound? President's address, 1960. AM. J. ROENTGENOL., RAD. THERAPY & NUCLEAR MED., 1960, *84*, 809–812.

* From the Department of Radiology, Harper Hospital, and Wayne State University, Detroit Michigan. Presented at the Sixty-second Annual Meeting of the American Roentgen Ray Society, Miami Beach, Florida, September 26–29, 1961.

logic organization the chronologic sequence of these trends and through its official JOURNAL has realistically expressed their impact on the Specialty as a whole, by scrupulously guarding at the same time the principle of Integrated Radiology.

The early workers, among which were many members of this Society, by courage, determined efforts and unrelenting perseverance overcame the many adversities particularly inherent in the medical application of such lethal agents as the roentgen rays and radium. Many successes were achieved, and to quote Sir Thurston Holland[2] "If it was at the time a bit of a nightmare . . . one has forgotten the failures, but has not forgotten the successes."

A perusal of the *American X-Ray Journal* and the *Transactions of the American Roentgen Ray Society* published in those early days reveals that almost everything in our present day practice of radiology had been conceived and to some degree elaborated before 1906. The position of roentgenology as a separate "Department of Medical Science" was clearly recognized and Dr. Hickey in 1906 wrote in the first editorial of THE AMERICAN QUARTERLY OF ROENTGENOLOGY[3]: "The roentgenologist should maintain always the position of consultant, who gives diagnostic conclusions based upon special lines of investigation. In regard to treatment, he should not allow himself to be dictated to either as to the number or character of his treatments."

After this first heroic decade progress has occurred at a swift pace. It soon became evident, however, that the tremendous accumulation of radiologic knowledge could not be stored in one single mind and that the ever-broadening scope of this knowledge in the medical field could be better achieved by devoting special attention to a branch of knowledge for which a person is distinguished. Thus the idea of a specialist within the specialty originated and since the 1920's radiology has been practiced more and more in the form of two distinct disciplines: Diagnostic Roentgenology and Therapeutic Radiology. Diagnostic Roentgenology fully emerged from its position of a "unicellular plant" in some obscure basement room and in the words of Dr. Reynolds[4] "stands at the crossroads of the corridors of the hospital forming the keypoint from which knowledge is disseminated in many directions to the benefit of the patient and to the advance of medical progress." Therapeutic Radiology, encompassing the use of the roentgen rays and the radiations of some members of the earth-born radioactive families, in particular for treatment of cancer, likewise gradually grew, after many initial difficulties, into an extremely valuable force of medical science. In our country, the two Disciplines, excepting in some of the large institutes, were never completely separated from each other, although trends toward separation were noted from time to time.

Then new trends developed within both major Disciplines. The notable progress made in all phases of medicine has brought with it a greater concentration on the study and treatment of certain individual organs and systems of the human body, resulting in the creation of various subspecialties in several Disciplines other than Radiology. This new development often made it necessary for the radiologist to apply his broad fundamental knowledge and the technical methods he already acquired to specific, more limited, fields in an endeavor to become more proficient in the diagnosis, and, at times, treatment of the disease of that organ or system. The eagerness in accomplishing this important task sometimes transcended into a trend toward complete separation from the main Discipline. Sosman,[5] as early as 1940, sounded a word of warning against such a trend. "And I may

[2] THURSTAN HOLLAND, C. X-rays in 1896. *Brit. J. Radiol.*, 1938, *11*, 1–24.

[3] HICKEY, P. M. Editorial. AM. QUART. ROENTGENOL., 1906, *1*, 1. Republished in: AM. J. ROENTGENOL., RAD. THERAPY & NUCLEAR MED., 1956, *75*, 2.

[4] REYNOLDS, L. Role of radiology today, its responsibility and its import upon our life. AM. J. ROENTGENOL. & RAD. THERAPY, 1948, *60*, 443–447.

[5] SOSMAN, C. M. Specialization within roentgenology. AM. J. ROENTGENOL. & RAD. THERAPY, 1940, *44*, 645–648.

warn them for you" said Dr. Sosman "that they must at the same time maintain contact and continue their cooperation with other work in Radiology, lest they become one of those ultra-specialists who digs himself deeper and deeper into his narrow pit until he thinks the rim of his hole is the horizon of knowledge."

The concentration on the study and treatment of certain individual organs or systems has brought with it even more definite trends in the realm of Therapeutic Radiology. The early practice of the use of radium or radon as operative procedures on an empirical basis by nonradiologists has been continued in many places, even after Therapeutic Radiology has become a Discipline. The great progress made in achieving accurate dosimetry, in general, has been slowed down rather significantly through the difficulty of estimating dosages with such procedures and trying to integrate them in the over-all plan of irradiation, although endeavors to correct the situation are being made. The introduction or revival of very radical surgical procedures for the treatment of cancers of certain regions and the division into subspecialties for the performance of the procedures in these regions have created radiotherapeutic problems which could have disastrous results, unless there is the closest cooperation and understanding between members of the various Disciplines participating in the treatment.

The dramatic events of World War II have given a tremendous impetus to a third major Discipline of Radiology which now is called Nuclear Medicine. Since the start of the general distribution of radioactive isotopes of nearly all the elements by the Atomic Energy Commission in 1946, this Discipline has reached unexpected proportions. At the present time, radioactive isotopes are being employed both in the diagnostic and therapeutic fields.

In the diagnostic field these are utilized for uptake studies; dilution techniques; flow or diffusion tests, mostly in conjunction with the cardiovascular system; manifold biochemical and metabolic procedures; and various scanning procedures. Many of them form an indispensable part of the routine roentgenologic examination so that, in order to emphasize their great clinical significance, the name of Diagnostic Roentgenology has been changed to that of Diagnostic Radiology.

In the therapeutic field, for obvious reasons, the application has followed very much the pattern established on the basis of clinical experience gained with the use of earthborn natural radioactive substances, but the greatest progress has been made in the use of certain isotopes, as for example, cobalt 60, cesium 137 and iridium 192 as sources for external gamma-beam teletherapy. Moreover, due to the fact that many of the radioactive isotopes, unlike radium or radon, can also be employed in solution or with appropriate liquid vehicles, considerable expansion of the use of local tissue irradiation, especially in intracavitary and interstitial applications, has resulted. In addition, a new form of treatment, the so-called internal irradiation with radioactive isotopes, has been made possible.

Clearly, all three major Disciplines: Diagnostic Radiology, Therapeutic Radiology and Nuclear Medicine, have become important and inseparable parts of the Specialty of Radiology. Within these three major Disciplines, subspecialties may at times be necessary or advantageous. On the diagnostic side, one may enumerate the cardiovascular, neurologic, pediatric, urologic, orthopedic, gastrointestinal and other branches, especially in view of the amazing advance in the utilization of the special procedures. On the therapeutic side, one may include the regional therapeutic branches, as, for instance, the head and neck; gynecologic organs; breast; genitourinary system; lymphomas, in which chemotherapy singly or in combination with radiation therapy is still in an experimental stage; and others. Nuclear Medicine is divided and is an integral part of both the branches of Diagnostic and Therapeutic Radiology.

This principle of an Integrated Radiology, with its three major Disciplines and various subspecialties is amply reflected in the history of the official organ of our Society. After its organization in 1900, the proceedings of the Annual Meetings of the Society were published in the form of "*Transactions*." By 1906, however, following the overwhelming abundance of excitement and thrills of the first decade, leaving us a great heritage of triumph over nearly insurmountable difficulties, the Society has acquired such stature in the medical world that it was deemed appropriate to establish an official Journal. It was published first as a quarterly under the title of "The American Quarterly of Roentgenology," but its scope increased so rapidly that in 1913 it was changed to a monthly publication with the name of "The American Journal of Roentgenology."

Interest in therapy, with both roentgen rays and radium, was already considerable at this time and many articles on this subject were published in the Journal. Then in 1916 the American Radium Society was founded mainly with the purpose "to promote the scientific study of radium and other sources of ionizing radiation in relation to their physical properties and their therapeutic application." In 1922, the official Journal of our Society was also made the official Journal of the American Radium Society and in 1923 its title was changed to that of "The American Journal of Roentgenology & Radium Therapy."

One decade ago, at its Annual Meeting in Washington, D.C., in 1951, the American Roentgen Ray Society "recognizing the importance of radioisotopes and the rapidity with which they have been employed within recent years, not only in diagnostic procedures but also as therapeutic agents" decided to incorporate in the title of the Journal an additional term which "would connote the Journal's interest in atomic medicine." Since then the new title has been "The American Journal of Roentgenology, Radium Therapy & Nuclear Medicine."

Since 1956, with the celebration of its Golden Anniversary, the Journal has adopted the policy of grouping together articles pertaining to a certain subspecialty of Radiology, as mentioned above, and publishing them in a single issue. This policy enables comprehensive coverage of a chosen topic of the subspecialty but, by making use of any phase by the three major Disciplines, it broadens the horizon of our knowledge and permits us to keep pace with the enormous strides which are continuously being made in the unrelentless advancement of the Specialty of Radiology as a whole.

Thus Integrated Radiology has served us well. But Integrated Radiology carries the seeds of a future even more brilliant than its glorious past. Built, as it is, on the fundamentals of the Universe, it already embraces all aspects of the man-made radiations and the earthborn natural radiations and there is every indication that soon it will be extended to the space radiations, both man-made and spaceborn, for the immeasurable benefit of mankind.

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ARTERIOSCLEROTIC NARROWING OF RENAL ARTERIES ASSOCIATED WITH HYPERTENSION*

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OVER fifty years ago Janeway[15] showed that postoperative blood pressure persisted at a significantly high level for more than a month when the renal arteries of dogs were constricted by ligatures. In 1934, Goldblatt *et al.*,[12] by means of a clamp that they devised, constricted the renal arteries in dogs. The amount of constriction which was induced varied considerably and the extent of constriction was changed over a time interval of several weeks. Constriction of one renal artery was followed by a slight to moderate elevation of blood pressure, which then tended to return toward normal levels. Constriction of both renal arteries was followed by the production of systolic hypertension to a varying degree in all of the dogs. Since the time of that experiment, much work has been performed on the relationship of renal ischemia to malignant hypertension. Recently the relationship of arteriosclerotic narrowing of the renal arteries to hypertension has been studied.

This report is based on 1,332 aortograms which were made at the Methodist Hospital, Houston, Texas, between 1953 and 1960 and which were evaluated for renal artery narrowing.

PRESENT CONCEPT

The most plausible and widely accepted present day concept regarding the etiology of renovascular hypertension is that decrease in renal artery pulse pressure by renal artery narrowing leads to the partial conversion of pulsatile to continuous blood flow in the kidneys with anoxia of the tubular cells as a result. This leads to increase in cellular membrane permeability and the release of the large renin molecule in the tubules. This enzyme acts on a plasma substrate to form a decapeptid called hypertensin I. (This substance is not the active vasoconstrictor which produces the hypertension.) Hypertensin I is converted to hypertensin II and the latter substance is the active vasoconstrictor which has been purified and shown to be five times more active than adrenalin as a vasopressor. Hypertensin II is an octapeptid and its continuous formation produces the generalized systemic hypertension which results.

An arteriosclerotic plaque near the origin of one renal artery reduces renal arterial pulse pressure and can cause malignant hypertension. The hypertension affects the entire body, including the opposite unobstructed kidney. The involved hypotensive kidney is protected from the generalized hypertension by the arteriosclerotic plaque in its renal artery so that histologic nephrosclerotic changes do not occur in this kidney. Decreased renal blood flow, glomerular filtration and sodium and water excretion result. The urine/plasma ratio of inulin is increased. The unobstructed hypertensive kidney shows the reverse effect. Revascularization of the hypotensive kidney may remove the stimulus for the production of the hypertensive cycle. If both renal arteries are narrowed, both kidneys may be protected from the hypertension and revascularization of the kidneys will result in a marked decrease in the hypertension. The malignant nephrosclerotic

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changes will not have occurred in either kidney and a dramatic response to the surgical procedure can be expected.

AORTOGRAPHY

A translumbar aortic puncture is performed with the tip of the needle above the origin of the celiac axis. Then 10 to 25 cc. of 70 per cent urokon is injected, either manually or through an automatic injector, and the film is exposed near the end of the injection. The abdominal aorta and its many branches, including both renal arteries, are well shown and arteriosclerotic narrowing of the renal arteries, usually occurring at or near their origin in the abdominal aorta, can be demonstrated. Occasionally, diffuse arteriosclerotic disease of some of the peripheral renal arteries is evident and rarely aneurysmal dilatation of one of its branches is seen. Only 25 cc. of urokon is injected in any given day, since impaired renal function and even death can result from excessive amounts of contrast material.

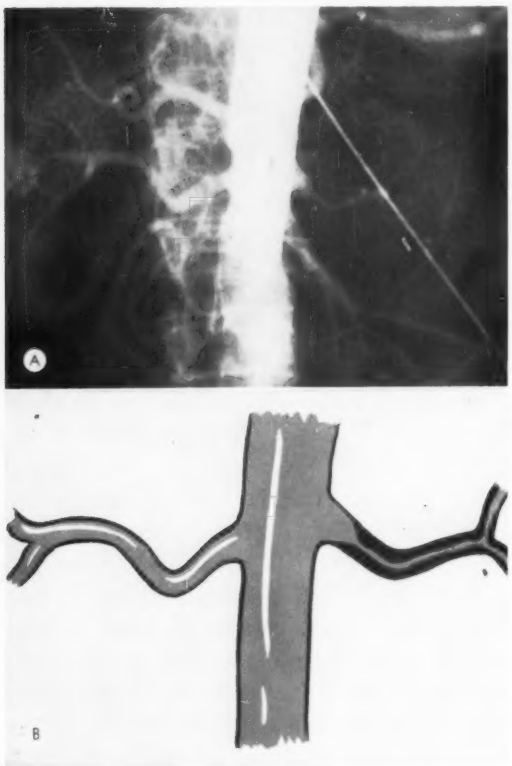

FIG. 1. Case I. (*A*) Aortogram showing diffuse narrowing of the left renal artery. The right renal artery is not significantly narrowed. (*B*) Drawing illustrating left renal artery narrowing.

ILLUSTRATIVE CASES

CASE I. B.S., a white male, seventy-six years old, had had moderate hypertension for twenty years and a known abdominal aortic aneurysm at the time of admission. Aortography demonstrated diffuse narrowing of the left renal artery (Fig. 1, *A* and *B*). An excretory pyelogram revealed impaired function of the left kidney. The abdominal aortic aneurysm was resected and replaced with a graft. Although, the left kidney was noted at surgery to be atrophic, no operative procedure was performed on it or on the left renal artery. The patient has remained well since the operation but the moderate hypertension has persisted.

CASE II. F.S., a white male, sixty-eight years old, had an eight months' history of frontal and occipital headaches and hypertension. Renogram showed vascular insufficiency on the left. The aortogram (Fig. 2, *A* and *B*) disclosed stenosis of the left renal artery just proximal to its bifurcation with poststenotic dilatation. Endarterectomy with patch grafting was performed on the left renal artery and a postoperative aortogram (Fig. 2, *C* and *D*) showed successful removal of the arteriosclerotic plaque in the left renal artery. The patient's postoperative blood pressure was normotensive.

CASE III. J.M., a white male, fifty-two years old, had a one year history of hypertension and headaches. He presented with an enlarged heart and generalized arteriosclerosis. The aortogram (Fig. 3, *A* and *B*) demonstrated right renal artery stenosis and an abdominal aortic aneurysm. The aneurysm was resected and a graft extending from the abdominal aorta to both common iliac arteries with a limb up to the right renal artery was inserted (Fig. 3, *C*, *D* and *E*). The patient was normotensive postoperatively.

CASE IV. E.M., a white female, forty-nine years old, gave a history of progressive hypertension of two years' duration with headaches, scotomata and syncopal episodes. The blood pressure ranged from 190/90 to 240/125 mm. of mercury. Split renal function studies showed a slight reduction of sodium and water excretion and of inulin and para-aminohippuric acid

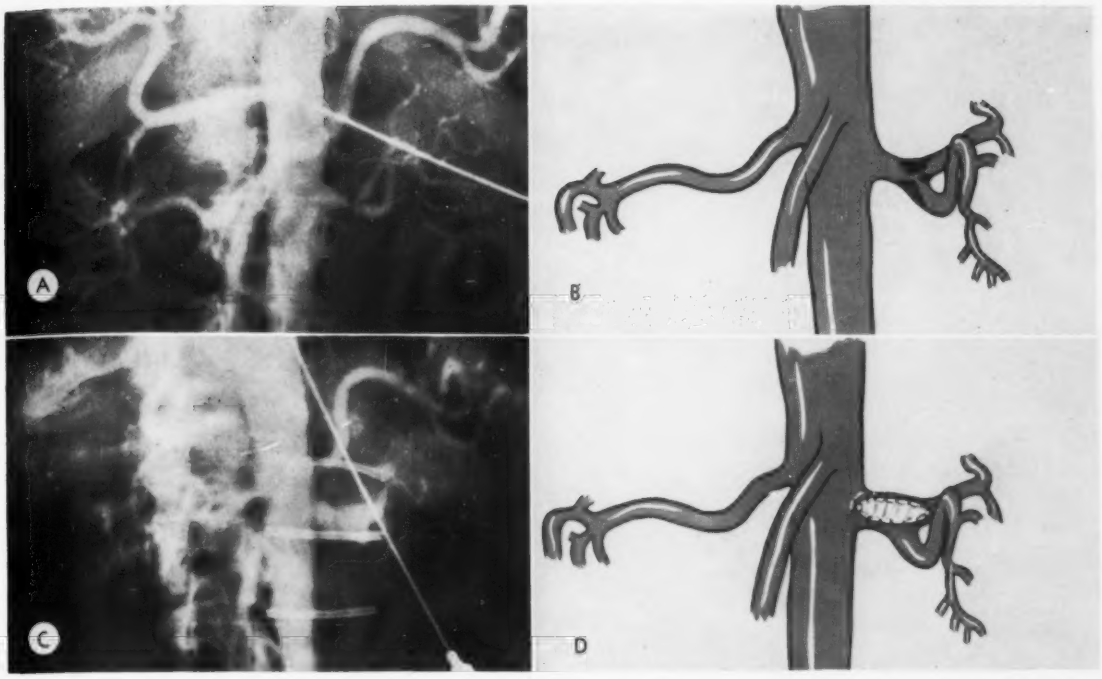

Fig. 2. Case II. (*A*) Aortogram demonstrating narrowing of the left renal artery; note also the early bifurcation. (*B*) Drawing illustrating the renal artery narrowing. (*C*) Postoperative aortogram showing the results of endarterectomy and patch grafting. (*D*) Drawing of the patch graft.

clearance in the right kidney. The aortogram (Fig. 4, *A* and *B*) revealed stenosis of the right renal artery. A bypass graft was inserted which extended from the abdominal aorta to the right renal artery (Fig. 4*C*). Postoperatively, the patient's blood pressure was in the normotensive range.

Case V. L.S., a white female, forty-eight years old, had a blood pressure of 180/90 to 220/120 mm. of mercury; her hypertension was first discovered six weeks before hospitalization. The aortogram (Fig. 5) demonstrated lower pole accessory renal arteries bilaterally with slight narrowing and poststenotic dilatation in the accessory right renal artery. There was a pulse pressure gradient in the accessory right renal artery. The patient underwent patch graft angioplasty to the accessory right renal artery and after operation she was normotensive.

Case VI. H.R., a white male, fifty-seven years old, had moderate hypertension of three years' duration with increasing claudication in both legs during that period. The aortogram (Fig. 6) demonstrated complete occlusion of the abdominal aorta just below the level of the renal arteries. Aortic-femoral bypass grafts were inserted and the patient had complete relief of the claudication; however, the moderate hypertension persisted. An occluded lower pole accessory right renal artery was suspected from the aortographic findings.

Case VII. A.N., a white male, forty-three years old, had symptoms of malignant hypertension of two months' duration. The blood pressure ranged from 190/100 to 280/150 mm. of mercury. Hypertensive retinopathy had produced almost complete blindness. An excretory pyelogram revealed delayed excretion by the right kidney. An aortogram (Fig. 7*A*) demonstrated stenosis of the right renal artery. An aortic-iliac bypass procedure was performed with a limb extending up to the right renal artery (Fig. 7*B*). Postoperatively, the patient was normotensive; the hypertensive retinopathy subsided and vision was normal within three weeks.

Case VIII. W.D., a white male, fifty-nine years old, was referred because of an abdominal aneurysm. The patient had moderately severe hypertension. An aortogram (Fig. 8*A*) revealed stenosis of the proximal portions of each renal artery. The abdominal aortic aneurysm was

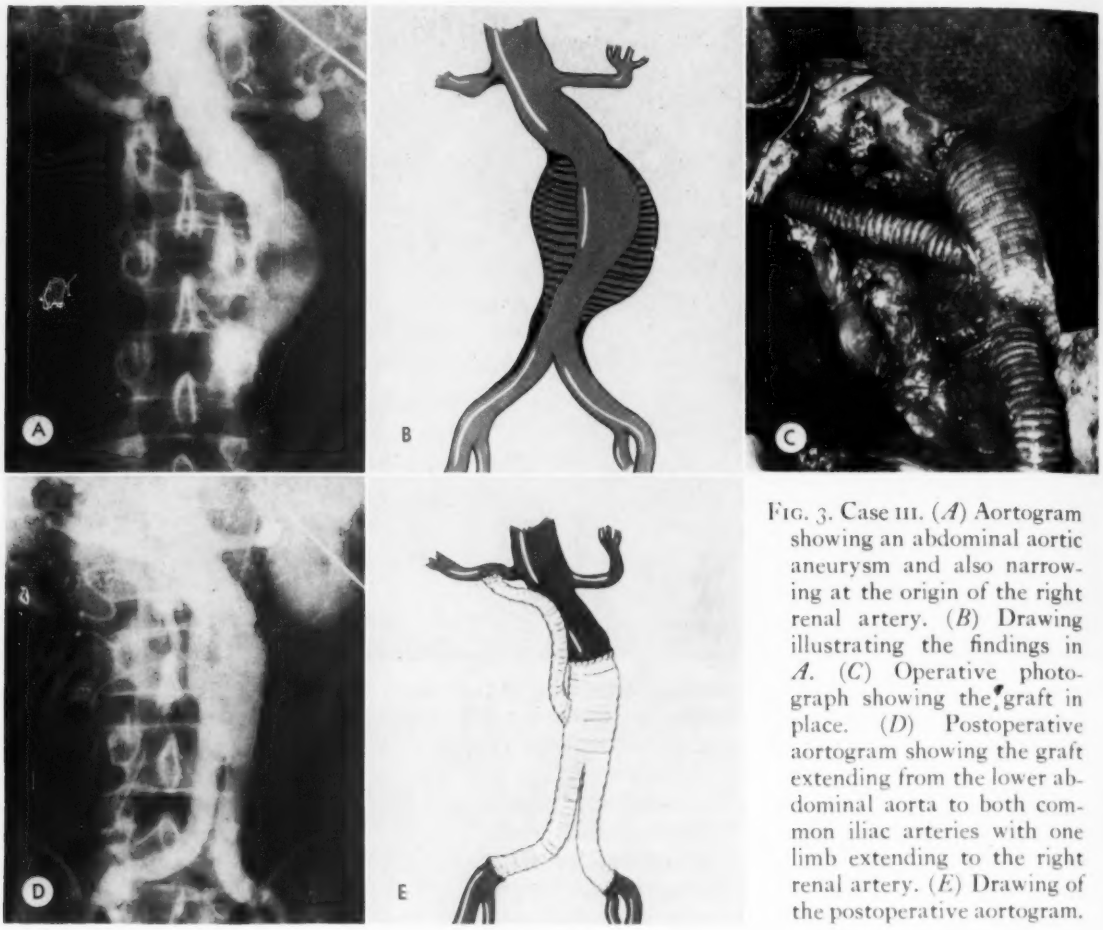

Fig. 3. Case III. (*A*) Aortogram showing an abdominal aortic aneurysm and also narrowing at the origin of the right renal artery. (*B*) Drawing illustrating the findings in *A*. (*C*) Operative photograph showing the graft in place. (*D*) Postoperative aortogram showing the graft extending from the lower abdominal aorta to both common iliac arteries with one limb extending to the right renal artery. (*E*) Drawing of the postoperative aortogram.

resected and replaced by a dacron graft and limbs from the graft were extended to each renal artery (Fig. 8*B*). Postoperatively, the patient had moderate reduction of the hypertension.

Case IX. L.H., a white male, fifty years old, had had resection and homograft replacement of the aortic bifurcation in 1953 for occlusive disease. Following this, the patient had increasing hypertension for two years. The blood pressure range was 200/90 to 240/130 mm. of mercury. An aortogram (Fig. 9*A*) revealed narrowing of the left renal artery and a graft was inserted extending from the abdominal aorta homograft to the left renal artery (Fig. 9*B*). Postoperatively, the patient had a blood pressure range of 118/80 to 160/85 mm. of mercury.

Case X. C.T., a white female, fifty-five years old, had hypertension of six years' duration with repeated black-out spells, numbness of the left arm and claudication of the left leg. After a right carotid endarterectomy was performed, an aortogram (Fig. 10) revealed narrowing of the right renal artery and of the lower pole accessory left renal artery. Resection and graft replacement of the aortic bifurcation with a patch graft angioplasty of the right renal artery were done. The patient has remained asymptomatic since the operation, although mild hypertension has persisted. The latter is probably explained on the basis of the narrowed accessory left renal artery.

Case XI. J.H., a white female, fifty-seven years old, had hypertension of five years' duration with progressive claudication for three years. The blood pressure range was 160/90 to 220/120 mm. of mercury. An aortogram (Fig. 11*A*) revealed narrowing of the left renal artery at its origin and an excretory pyelogram (Fig. 11*B*) showed impaired function of the left kidney. An aortic-femoral bypass graft was in-

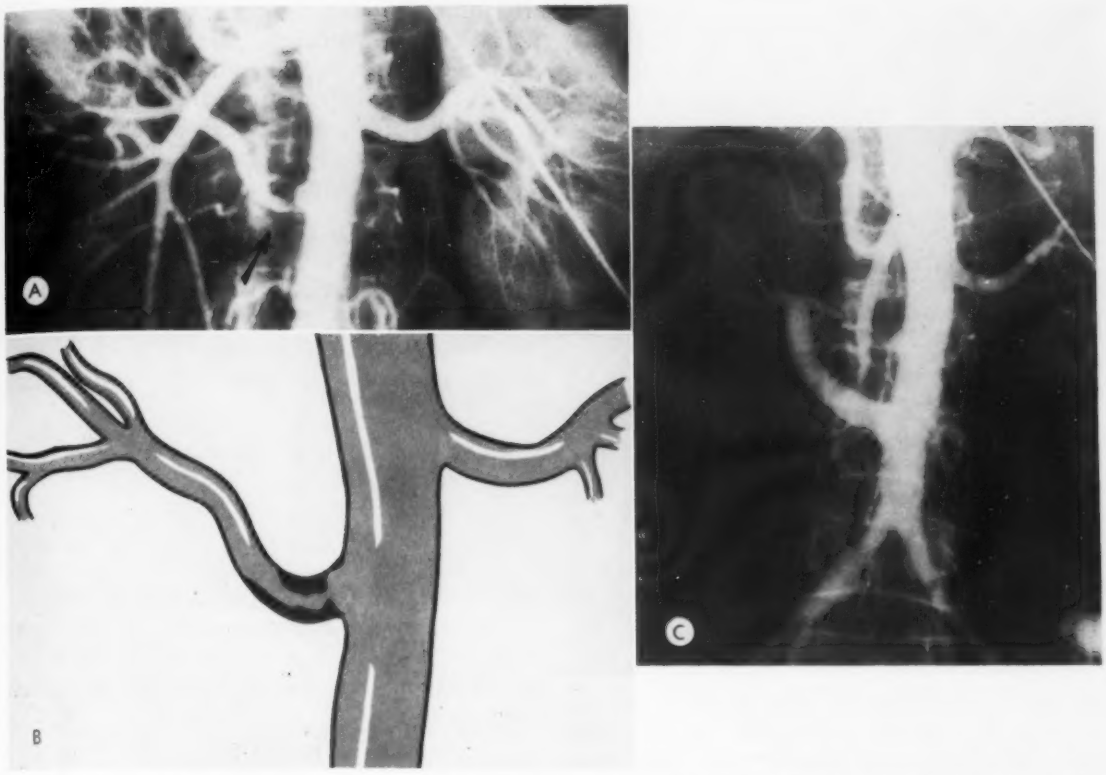

FIG. 4. Case IV. (*A*) Aortogram showing narrowing of the right renal artery (arrow). (*B*) Drawing illustrating narrowing of the right renal artery. (*C*) Postoperative aortogram showing the limb of the graft extending to the right renal artery.

serted bilaterally with a limb extending up to the left renal artery. The blood pressure postoperatively remained within the normotensive range and there was no longer any claudication.

CASE XII. H.G., a white male, sixty-four years old, had hypertension and a history of right hemiplegia with subsequent recovery. An aortogram (Fig. 12) revealed moderate narrowing of the left renal artery at its origin. A left renal artery bypass graft was inserted as well as aortic-iliac grafts and the patient was normotensive postoperatively.

CASE XIII. H.W., a white male, fifty-five years old, had hypertension of several years' duration with progressive claudication of both legs for two years. The blood pressure range was 210/110 to 260/120 mm. of mercury. The

FIG. 5. Case V. Aortogram showing lower pole accessory renal arteries bilaterally. There is slight narrowing with poststenotic dilatation on the right.

FIG. 6. Case VI. Aortogram illustrating the usual location for complete abdominal aortic occlusion. An occluded anomalous renal artery to the lower pole of the right kidney was suspected.

renogram revealed decreased vascularity of the left kidney and an aortogram (Fig. 13) showed stenosis of the left renal artery. A bilateral aortic-femoral bypass graft with a limb extending up to the left renal artery was inserted and the patient's blood pressure gradually declined to 140/80–160/90 mm. of mercury.

CASE XIV. W.B., a white male, forty-nine years old, had a three year history of hypertension and dizziness. An aortogram (Fig. 14) revealed stenosis of the right renal artery. An aortic-femoral right renal bypass graft was inserted and the patient was normotensive postoperatively.

CASE XV. R.S., a white male, forty-nine years old, had claudication in the right leg for two years with no history of hypertension. The aortogram (Fig. 15) revealed bilateral accessory renal arteries with slight narrowing at the origin of the right lower renal artery. The patient underwent endarterectomy of the right common iliac artery with complete relief of the claudication. His blood pressure remained normal.

RESULTS

A three-man team reviewed 1,332 lumbar aortograms and evaluated the appearance of the renal arteries. Most of the patients

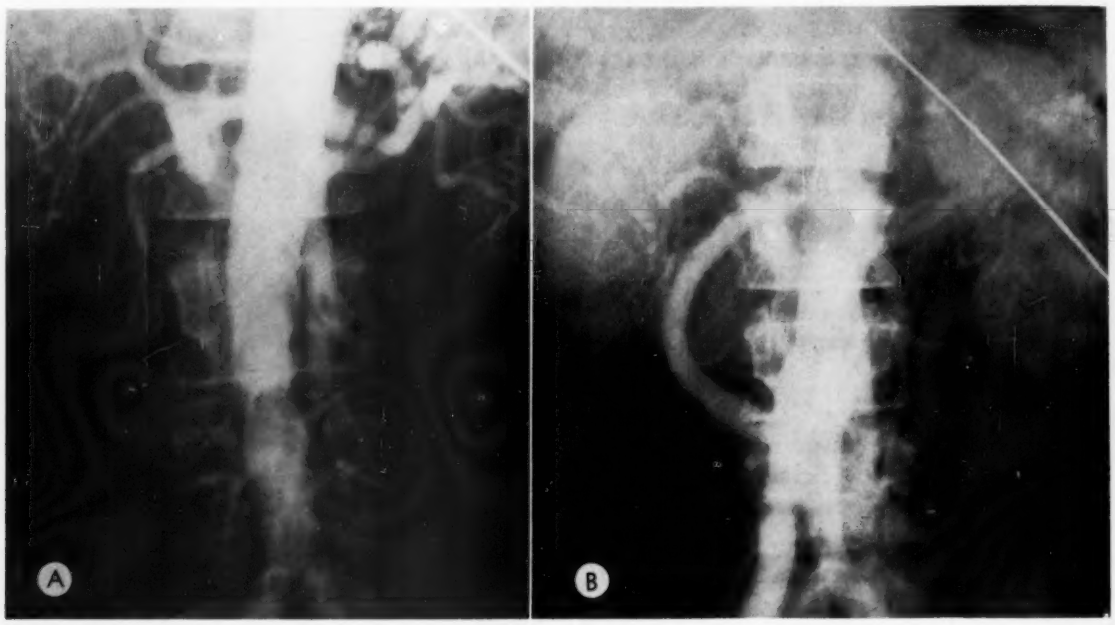

FIG. 7. Case VII. (*A*) Aortogram illustrating almost complete occlusion of the right renal artery. (*B*) Postoperative aortogram showing the limb of the graft extending to the right renal artery.

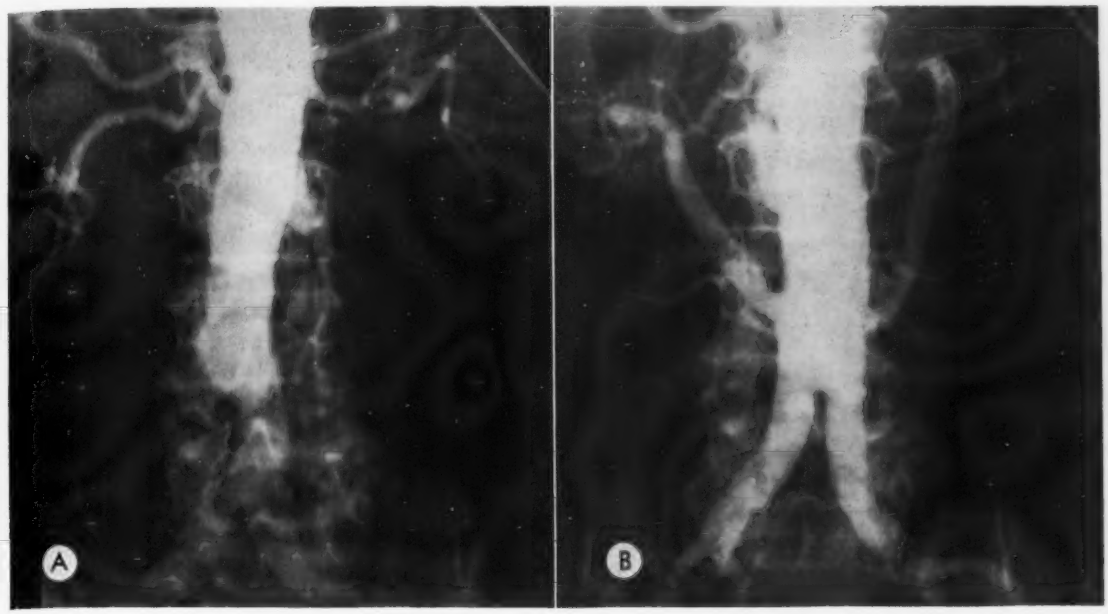

Fig. 8. Case VIII. (*A*) Aortogram demonstrating narrowing of both proximal renal arteries. (*B*) Postoperative aortogram showing a limb of the graft extending to each renal artery.

had aortography performed because of aneurysm or occlusive disease in the abdominal aorta or because of circulatory impairment to the lower extremities. In other words, most of these cases had advanced arteriosclerotic disease as the cause of their presenting complaint and relatively few of the examinations were done primarily for the evaluation of hypertension. Renal artery anomalies were noted and kidney size and renal function were estimated by means of a follow-up pyelograms which, in most cases, had been taken ten minutes following the aortogram. Arteriosclerotic disease of the abdominal aorta and other branches was also noted. (This was found to be present in a very high percentage of the cases.) The age

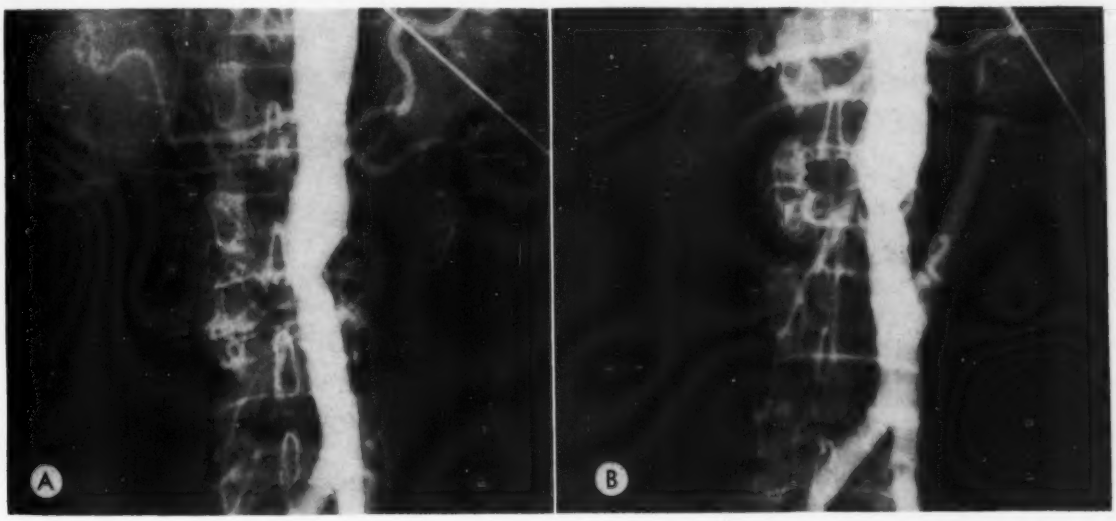

Fig. 9. Case IX. (*A*) Aortogram demonstrating stenosis of the left renal artery. (*B*) Postoperative aortogram showing the graft to the left renal artery.

FIG. 10. Case x. Aortogram made after a low aortic injection and showing stenosis of the right and inferior accessory left renal arteries. (This type of needle placement has been discarded because it can cause intramural dissection in a diseased aorta and renal artery occlusion may follow.)

range of the patients was eighteen to eighty-six years. The average age was fifty-five years.

Of the 1,332 aortograms, 437 were judged technically unsatisfactory for this study, since the renal arteries were not adequately visualized. Motion of the patient, improper centering of the film, excess dilution of contrast material, extravasation of contrast material, superimposition of other arteries over the renal vessels and direct injection into the celiac axis or superior mesenteric artery were some of the causes for inadequate demonstration of the renal arteries; however, most of these 437 studies were unsatisfactory because the aortic puncture was below the level of the renal arteries. (Injection at this level is discouraged at the present time because of the possibility of producing a dissecting aneurysm at the site of the needle puncture, since arteriosclerotic changes are usually more severe in the lower abdominal aorta than in the upper.) Also, the aortograms of 74 patients who had no blood pressure readings recorded on their hospital charts were not considered. This left a total of 821 aortograms which were used in the present series

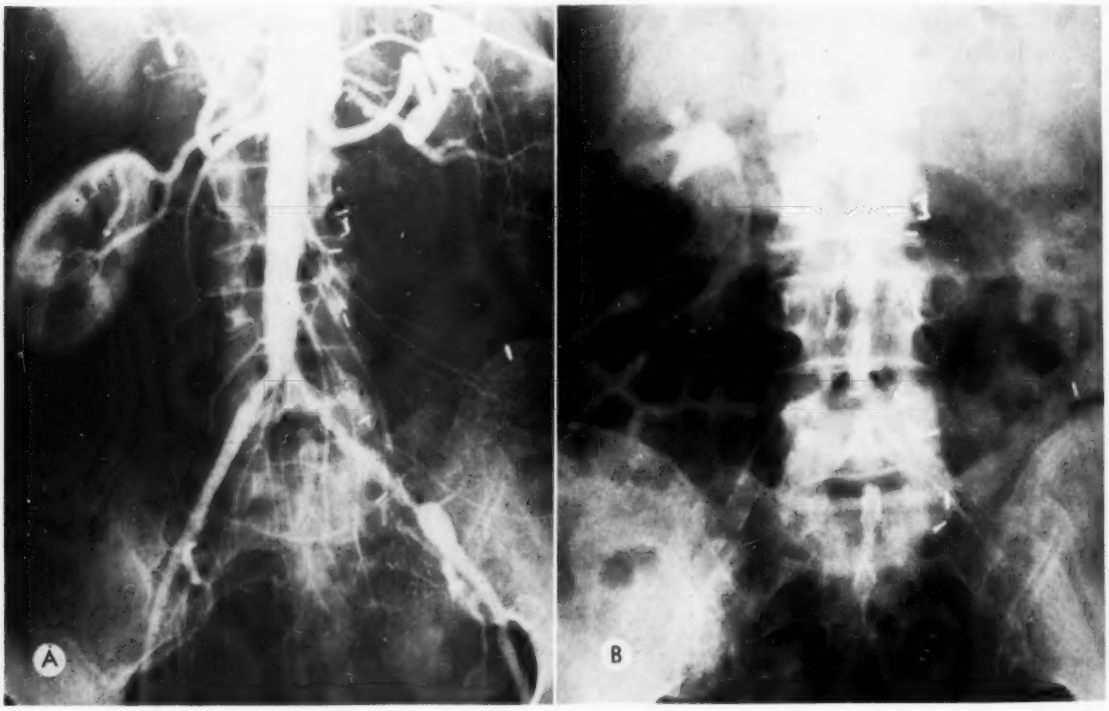

FIG. 11. Case xi. (*A*) Aortogram showing stenosis of the left renal artery. (*B*) Follow-up excretory pyelogram revealing impaired function of the left kidney.

Fig. 12. Case xii. Aortogram demonstrating stenosis of the left renal artery (arrow).

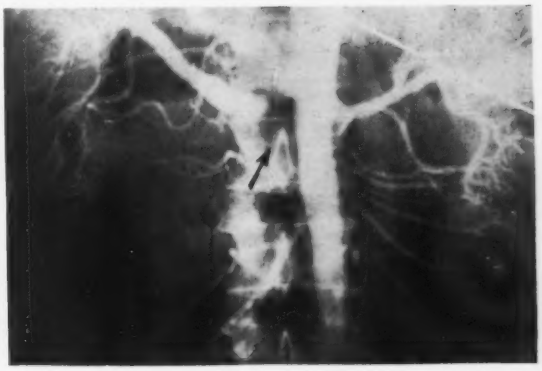

Fig. 14. Case xiv. Aortogram demonstrating stenosis of the right renal artery (arrow).

for the statistical evaluation of arteriosclerotic disease of the renal arteries and its relationship to hypertension (Table i).

The criteria for hypertension are, of course, a very important aspect of this statistical analysis and a slight change in the criteria will make a significant difference in the statistics.

The blood pressure readings were taken from the hospital charts and usually totaled six recordings. The first two, the last two, and the highest two readings were tabulated. Table ii shows the results when, as criteria for hypertension, two of the six blood pressure recordings showed a systolic or diastolic hypertension, or when there was one systolic and one diastolic elevation. These criteria were purely arbitrary and do not constitute a very strict definition. Many of the patients, however, were on medication for the treatment of hypertension. On the other hand, of course, errors can be made in the taking of the blood pressure.

Any diastolic recording over 90 mm. of mercury was considered hypertensive at any age. The baseline for the systolic hypertensive level was taken from insurance

Table I

Material

Total aortograms reviewed	1,332
Aortograms with inadequate visualization of renal arteries	437
Cases with no blood pressure readings recorded	74
Aortograms and cases used	821

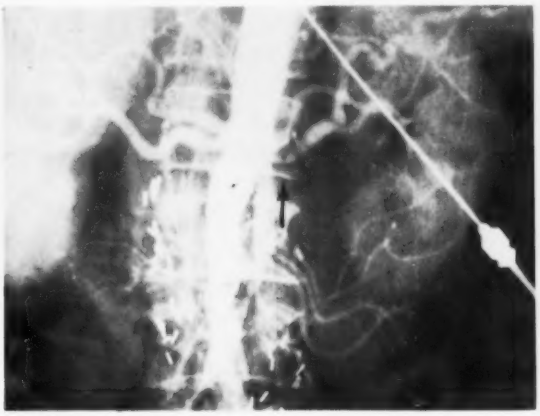

Fig. 13. Case xiii. Aortogram demonstrating stenosis of the left renal artery (arrow).

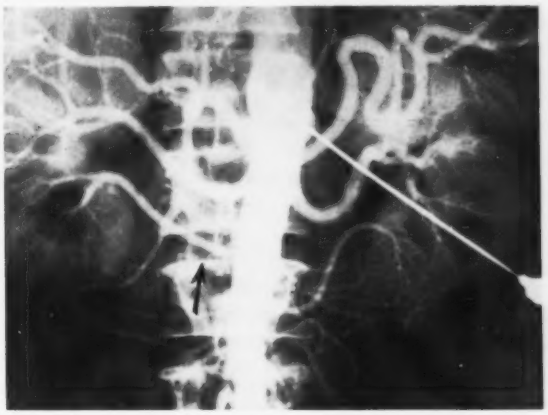

Fig. 15. Case xv. Aortogram demonstrating bilateral accessory renal arteries with stenosis evident on the right (arrow).

Table II

RENAL ARTERY STENOSIS AND HYPERTENSION*

(821 cases)

	No. of Cases	Per Cent
Hypertension	643	78.3
No hypertension	178	21.7
Renal artery stenosis (unilateral or bilateral)	416	50.7
Hypertension and renal artery stenosis	358	43.7
Arteriosclerotic occlusive disease (aortic, iliac, femoral or popliteal arteries)	737	89.7

* Criterion for hypertension; two of the six blood pressure readings showing a systolic or diastolic elevation; or one systolic and one diastolic elevation.

company statistical analyses of normal levels of blood pressure in various age groups. Under forty years of age, 140 mm. of mercury was considered the dividing line between normotension and hypertension. For each succeeding decade after forty, 5 mm. of mercury is added to give the upper limits of normotension in the population of the United States as a whole.

Table II shows that 78.3 per cent of the total 821 patients had hypertension; 50.7 per cent had narrowing of one or both renal arteries demonstrated by aortography, and 43.7 per cent had both hypertension and renal artery narrowing. Unilateral narrowing was slightly more common than bilateral and the right artery was stenosed slightly more frequently than the left (Table III). Associated arteriosclerotic disease (in the abdominal aorta, iliac, femoral or popliteal arteries) was present in almost all the cases (89.7 per cent).

A ten minute excretory pyelogram was usually made following the aortogram and this was evaluated for evidence of impaired renal function. Many authors describe absent or delayed excretion of contrast material as a sign of impaired blood supply to a kidney and some consider that a definitely smaller kidney on one side indicates impaired renal function. These signs, when present, were usually found to be in the patient who had aortographic evidence of narrowing of a renal artery. However, in the overwhelming majority of the cases, excretion of contrast material and kidney size were normal (Table IV). It appeared to us that excretory pyelography following aortography adds very little to the evaluation of the blood supply to the kidneys.

Each renal artery in the 821 aortograms was judged as normal, questionably diseased, showing definite evidence of arteriosclerosis, or inadequately visualized for technical reasons.

The accessory renal arteries were not tabulated but it was our general impression that they are frequently present, and

Table III

RENAL ARTERY STENOSIS AND HYPERTENSION*

(821 cases)

	No. of Cases			Per Cent
Renal artery stenosis (unilateral or bilateral)	416			50.7
Hypertension		358		43.7
Bilateral renal artery stenosis			155	18.9
Unilateral renal artery stenosis			203	24.8
Right renal artery stenosis			107	13.1
Left renal artery stenosis			96	11.7
No hypertension		58		7.0
Normal renal arteries, bilaterally	235			28.7
Hypertension		154		18.8
No hypertension		81		9.9

* Same criterion for hypertension as in Table II.

TABLE IV

RENAL ARTERY STENOSIS AND HYPERTENSION*
(821 cases)

	No. of Cases	Per Cent
Associated arteriosclerotic disease (aortic, iliac, femoral or popliteal arteries)	737	89.7
Nonvisualization of a kidney on the pyelogram	2	0.2
With hypertension and renal artery stenosis	1	0.1
Contracted kidney	8	1.0
With hypertension and renal artery stenosis	6	0.7
Delayed excretion	62	7.5
With hypertension and renal artery stenosis	46	5.6

* Same criterion for hypertension as in Table II.

as many as three renal arteries to a kidney were seen. A low accessory renal artery frequently is partially or completely occluded and many times an occluded renal artery not visualized on the aortogram can be suspected because of a relatively avascular area (usually the lower pole) seen on the nephrogram.

When the criteria for hypertension were varied slightly there was a significant effect on the statistics. A second statistical analysis of the association of arteriosclerotic narrowing and hypertension was made when, as the criterion for hypertension, three of the six systolic or two of the six diastolic recordings were over the limit for the age of the patient (Table v). Of the total 821 cases, 54.5 per cent had hypertension and 33.7 per cent had both hypertension and renal artery stenosis.

CONCLUSIONS

Of the total 821 cases, 43.7 per cent had both renal artery narrowing and hypertension (Table II). Arteriosclerotic disease of other arteries was almost always present. Delayed or absent excretion of contrast material by a kidney or the presence of a contracted kidney was not evident with any significant frequency. When more stringent hypertension criteria were utilized, 33.7 per cent of the 821 cases had both hypertension and renal artery disease (Table v).

TABLE V

RENAL ARTERY STENOSIS AND HYPERTENSION*
(821 cases)

	No. of Cases	Per Cent
Hypertension	447	54.5
No hypertension	374	45.5
Renal artery stenosis	417	50.7
Hypertension and renal artery stenosis	276	33.7

* Criterion for hypertension: three of the six blood pressure readings showing a systolic elevation; or two diastolic elevations.

SUMMARY

In a review of 1,332 aortograms, 821 were found satisfactory for evaluation of the renal arteries. The hospital charts of the 821 cases were studied and blood pressure readings were recorded. The statistical relationship of renal artery narrowing to hypertension was tabulated.

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ANALYSIS OF THE RADIO-RENOGRAM CURVE*

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THE use of the I^{131} renogram as a test for kidney function has been described.[4,6] The chief application of this procedure is as a screening test to rule out unilateral renal disease in hypertensive persons,[5] and to detect gross abnormalities in renal function that involve large portions of kidney parenchyma.[1] The major limitation in its use is that this test is essentially qualitative and subject to individual interpretation.[2]

The work presented here proposes a method of analysis of the radio-renogram that permits a more objective interpretation.

METHOD

Patients are placed on a bed in a prone position. The anatomic location of the kidneys is determined beforehand by abdominal roentgenography. A 2 inch sodium iodide (Tl) scintillation crystal in a lead collimated probe is placed over each kidney. A third scintillation detector is placed over the posterior thorax at the level of the heart. Each detector is connected to a count rate meter and then to a rectilinear strip chart recorder.

One microcurie of I^{131} labeled hippuric acid per 3 kg. of body weight is injected intravenously. The levels of radioactivity over each kidney and the heart are recorded.

Control renograms were made of 50 healthy university students. Renograms were also made of 22 hospital patients with hypertension. One or more of the standard renal function tests, including intravenous pyelography, phenolsulfonphthalein excretion, aortography, differential clearance studies, and blood and urine chemistries were also performed. On the basis of these clinical tests, these patients were found to have no renal dysfunction associated with their hypertension. In addition, renograms were made of a group of 9 patients with clinically proved renal disease.

RESULTS

A normal hippuric acid renogram is shown in Figure 1. Characteristically, it has the three segments described by Taplin *et al.*[3] The first, or vascular, segment is shown by the portion of the curve labeled *AB*. The second, or concentration, phase is *BC*. The third, or excretory, segment is shown by *CEF; F* being the point at which a new base line or equilibrium is reached.

In order to make the analysis for this study, each renogram was prepared as follows: from point *B* a horizontal line was drawn to intersect *CF* at point *E*. Vertical lines were then drawn from points *B*, *C*, and *E* to the base line. The time elapsed during the vascular segment of the curve is represented by t_b; the duration of the concentration segment is t_c; and t_e is the time during which the curve returns to a value equal to the height of the vascular peak.

To achieve standardization of the renograms the following considerations were made.*

1. "*X*" is equal to the maximum blood level of I^{131}.
2. "*Y*" is the peak of the concentration phase of the renogram curve.
3. Then $100(Y-X)/X$ is the percentage of the maximum blood level concen-

* For the purposes of this study and the development of arbitrary expressions of kidney function, the mathematic assumptions made are adequate. The authors acknowledge the fact that an accurate quantitative analysis of the renogram curve would involve considerations more complex than the linear relationships expressed herein.

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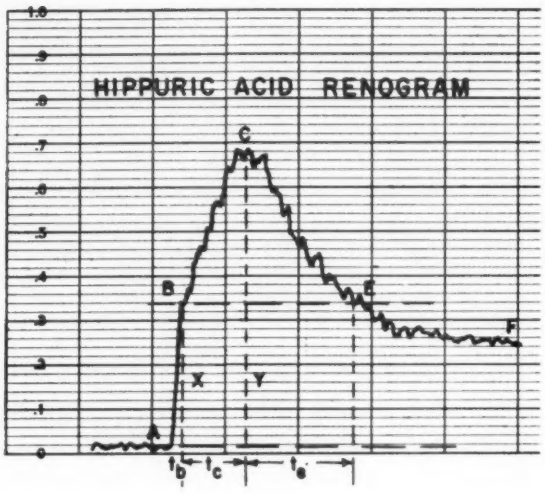

FIG. 1

trated by the kidney, or total concentration.

4. Total concentration divided by time t_c equals the percentage concentrated per minute, or minute concentration.
5. Total concentration divided by time t_e equals the percentage excreted per minute, or minute excretion.

The renograms are divided into three groups. Group I is the control group. Group II consists of the hypertensive patients with no demonstrated kidney disease. Group III is composed of those patients with unilaterally or bilaterally abnormal kidney function (demonstrated by tests other than the renogram).

The total concentration, minute concentration, and minute excretion values were determined for the left and right renogram curves of each person tested. The mean values and standard deviations of these determinations for the renograms in Groups I and II are shown in Table I and II.

Table III lists the results of the renograms made of the patients in Group III; that is, those with abnormal kidney function.

DISCUSSION

From the data in Table I it can be seen that, in the control group, there is no significant statistical difference in the normal values between the left and right renogram curves.

Similarly, a comparison of the renograms in the second group also shows no statistical difference between the values for the left and right renogram curves.†

In comparing the total concentration and minute concentration of Groups I and II with each other, it is seen that, as a whole, the hypertensive group has significantly lower levels ($p < .01$) of these two parameters than the control group. This disparity is more evident when comparisons between the minute excretion values of these two groups are made ($p < .005$). When considered individually, however, the majority of the hypertensive patients fall within the normal limits as defined by the control

† This study was originally undertaken using I^{131} diodrast as the test agent, and a wide divergency of normal values was found between the left and right renograms, presumably due to the concentration of diodrast in the liver. For this reason we have abandoned this compound in favor of I^{131} hippuric acid.

TABLE I

GROUP I—NORMAL CONTROLS

	Total Concentration (per cent)		Minute Concentration (per cent)		Minute Excretion (per cent)	
	Mean	Standard Deviation	Mean	Standard Deviation	Mean	Standard Deviation
Left Kidney	72.1	27.1	20.6	8.9	17.4	9.1
Right Kidney	73.7	31.8	20.0	9.8	16.9	9.0
Average of Both Kidneys	72.9	29.5	20.3	9.3	17.1	9.0

Table II

GROUP II—HYPERTENSIVE PATIENTS WITH NORMAL KIDNEY FUNCTION

	Total Concentration (per cent)		Minute Concentration (per cent)		Minute Excretion (per cent)	
	Mean	Standard Deviation	Mean	Standard Deviation	Mean	Standard Deviation
Left Kidney	54.9	26.8	17.7	7.2	11.0	7.1
Right Kidney	55.1	28.6	15.7	6.2	10.3	4.6
Average of Both Kidneys	55.0	27.7	16.7	6.7	10.6	5.8

group. As the number of normal and hypertensive patients in this study is increased, it may be possible to evaluate more fully the significance of this difference between the two groups.

Table III shows that the value of one or more of the three renogram parameters for the patients in the third group falls outside the normal limits given by the control group.

Table III

GROUP III—PATIENTS WITH ABNORMAL KIDNEY FUNCTION

Patient	Kidney	Total Concentration (per cent)	Minute Concentration (per cent)	Minute Excretion (per cent)	Clinical Diagnosis
1	Left	60.0	18.9	3.6	Chronic glomerulonephritis
	Right	27.5	8.7	2.4	
2	Left	53.1	18.1	4.9	Chronic glomerulonephritis
	Right	80.2	26.0	5.9	
3	Left	88.0	2.7	0.0	Left hydronephrosis;
	Right	22.0	2.7	1.4	Nonfunction of right kidney
4	Left	314.0	18.8	0.0	Acute glomerulonephritis
	Right	104.0	9.0	0.0	
5	Left	148.0	11.3	5.5	Chronic glomerulonephritis
	Right	178.0	17.5	7.4	
6	Left	167.5	4.9	0.0	Left hydronephrosis;
	Right	0.0	0.0	0.0	Nonfunction of right kidney
7	Left	14.9	8.1	9.4	Chronic pyelonephritis
	Right	23.8	10.6	6.8	
8	Left	168.0	9.6	0.9	Left hydronephrosis;
	Right	102.0	22.8	9.6	Normal right kidney
9	Left	106.0	5.5	0.0	Bilateral hydronephrosis
	Right	16.7	1.1	0.0	

CONCLUSION

A method for analysis of the radio-renogram is proposed. This method eliminates certain variables inherent in this procedure and permits a more objective interpretation of the renogram curve.

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EXTRARENAL CALYCES: THEIR RELATIONSHIP TO RENAL DISEASE*

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A RARE congenital anomaly of the renal pelvis and calyces, which has evoked little interest in the past, has been encountered with increasing frequency due to the awareness and recognition of this abnormality. Although it may appear as an incidental finding at postmortem examination, more often it has been found associated with renal disease. Reports describing extrarenal calyces have been sparse in the urologic literature, and our series of 8 cases suggests that this anomaly may occur more commonly than the literature would indicate.

CASES REPORTED IN THE LITERATURE

Eisendrath[2] presented a case of calculous hydronephrosis with extrarenal calyces and discussed 3 previously reported cases. The first of these, described by Fürstner, was a case in which the extrarenal calyces were present in a hypoplastic segment of a bifid kidney. The second case, mentioned by Herz, was one of bilateral extrarenal calyces. In the third case, reviewed by Apert, the extrarenal calyces entered the ureter without an intermediate pelvis. A case similar to that of Eisendrath was reported by Veseen.[3] In 1951, a sixth case, described by Dunn and Gibson,[1] showed ectopia and hydronephrosis associated with this congenital anomaly.

ANATOMY

The anatomic feature (Fig. 1) of this congenital anomaly is a discoid shaped kidney, flattened in the anteroposterior diameter, with the pelvis and the major and minor calyces located outside the renal parenchyma. The hilus is elliptical and

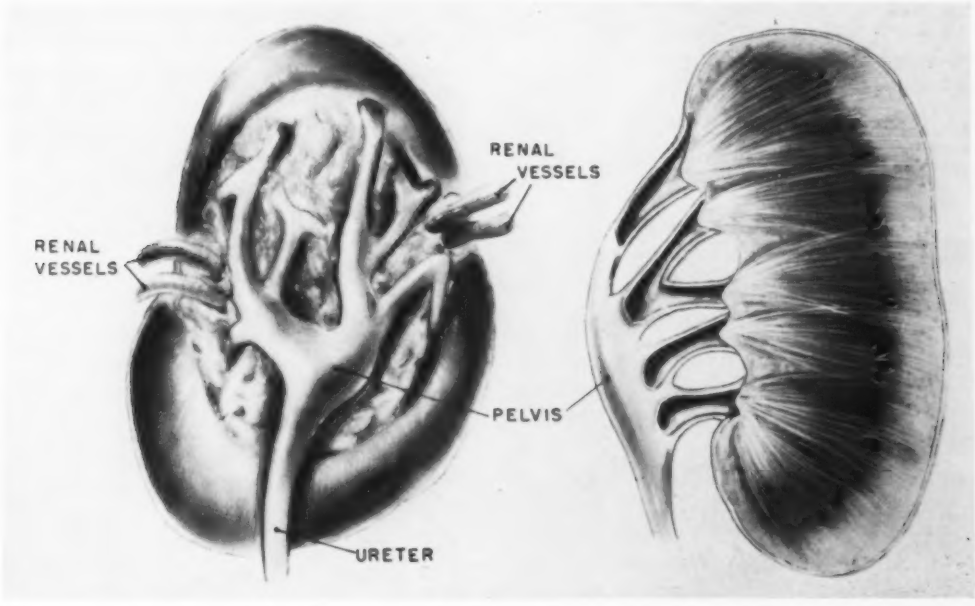

FIG. 1. Posterior aspect (left) and cross section (right) of an extrarenal calyceal system.

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covers most of the posterior surface, with the minor calyces attaching themselves to the kidney in this area. The cut section of the kidney reveals the usual appearance of cortical and medullary zones, with the papillae protruding into the minor calyceal cups, which are attached at the hilar surface. The renal vessels enter the kidney with an anomalous distribution, usually at the circumferential edge of the flat, widened hilus.

EMBRYOLOGY

The anomaly may be due to a disparity resulting from slow development of the nephrogenic anlage, or to a rapid development of the ureteric bud. At the end of the third or the beginning of the fourth week of gestation, the metanephros arises from the nephrogenic anlage and replaces the mesonephros in function. Simultaneously, the ureteric bud ascends in a cephalad direction toward the nephrogenic mass, which forms a cap around the blind end. As the growth continues, the blind end widens and invaginates, forming a cephalad and a caudad limb, representing the major calyces. Two more buds, a dorsal and a ventral, grow out of each limb, forming the four buds which constitute the major calyces; they are referred to as the collecting tubules of the first order. Each of these, in turn, produces two to four buds which develop into the minor calyces, or the collecting tubules of the second order. Should exaggerated or precocious development occur at this stage, the calyceal system could well develop prior to its coalescence with the nephrogenic mass. Conversely, a lag in the growth of the nephrogenic mass could delay its attachment to the collecting system, permitting extrarenal development of the first or second order of the collecting system.

DIAGNOSIS

This condition may be detected by excretory urography and retrograde pyelography when the renal pelvis is visualized outside the renal shadow. More characteristic is the pattern of the major calyces, which seem to flare out with a terminal incurvation, simulating the curved prongs of a small hand garden rake. When surgery is indicated, meticulous dissection of the renal hilus readily reveals the pelvis to be located in the perirenal tissues. The calyces can then be traced to their insertion in the renal parenchyma.

REPORT OF CASES

CASE I. A.C. This sixty-seven year old white male, because of continued urinary difficulties following a transurethral resection in 1946 at another hospital, had a second operative procedure in 1950 at the same hospital. The pathologic diagnosis of the tissue obtained at the second resection was adenocarcinoma of the prostate gland. Intravenous urography revealed a normal left kidney and an ectopic right kidney. On admission to the Veterans Administration Hospital in 1956 because of urinary symptoms, examination demonstrated a nonfunctioning right kidney and a normal left kidney, as well as generalized metastases. The patient expired in November, 1957. At autopsy, a typical extrarenal calyceal system of the right kidney was found. This anomaly had not caused any clinical symptoms.

CASE II. J.M. This sixty-two year old white male was admitted to the Flower Fifth Avenue Hospital in 1948 complaining of gross hematuria for the past eighteen years. This occurred at approximately six month intervals and would clear within twenty-four hours. Occasional dysuria was admitted, but no other symptoms were present. Physical examination and laboratory findings were essentially normal. The intravenous urogram and retrograde pyelogram revealed a left calculous hydronephrosis. The right side appeared normal. A left nephrectomy was performed and examination of the specimen (Fig. 2) showed extrarenal calyces attached to a discoid shaped kidney. The pelvis contained a large calculus causing urinary obstruction. The kidney exhibited advanced obstructive degeneration.

CASE III. R.B. This sixty year old white male was admitted to the medical service of the Veterans Administration Hospital suffering with diabetes, hypertension and amaurosis. Early signs of cardiac decompensation were present and during the work-up an intravenous urogram was obtained (Fig. 3*A*). This revealed

a right ectopic kidney with hydronephrosis. The urine showed 25–30 white blood cells per high power field and the patient was referred to the urology service for nephrectomy. The excised specimen (Fig. 3, *A* and *B*) revealed extrarenal calyces, pyelonephritis and arteriolo-nephrosclerosis. The hypertension was only slightly improved, decreasing from 232/118 to 186/92.

Case iv. J.L. This sixty-seven year old white male was admitted to the surgical service of the Veterans Administration Hospital for a possible colon malignancy. Physical examination revealed in the left upper quadrant a palpable mass which was associated with left flank pain radiating to the suprapubic area. The patient was referred to the urology service and an intravenous urogram and retrograde pyelogram demonstrated distortion of the calyceal system suggesting a renal tumor (Fig. 4, *A* and *B*). At surgery a large dark brown mass was found intimately adherent to the kidney. The kidney was removed and examination showed an organized perirenal hematoma of unknown origin. Dissection of the renal pelvis disclosed an extrarenal calyceal system. It was suggested that the perirenal hematoma may have been due to the rupture of an anomalous vein.

Case v. A.H. This fifty-three year old white male was treated on the medical service of the Veterans Administration Hospital for cardiac

Fig. 2. Case ii. Extrarenal calyces with impacted calculus in the pelvis.

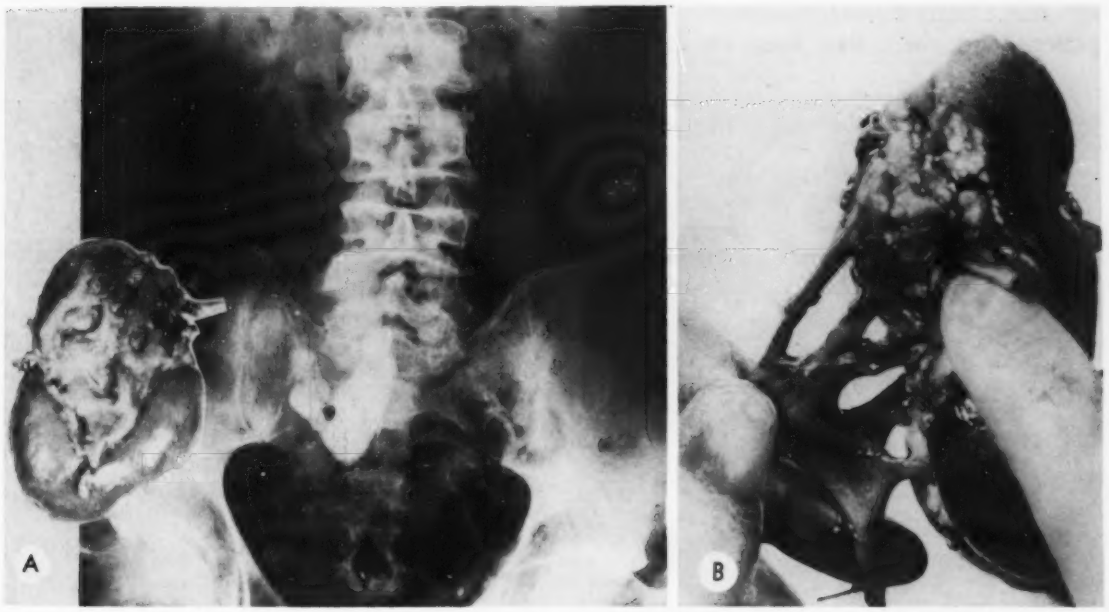

Fig. 3. Case iii. (*A*) Composite photograph of the pyelogram and excised kidney. (*B*) Lateral view showing the extrarenal location of the calyces.

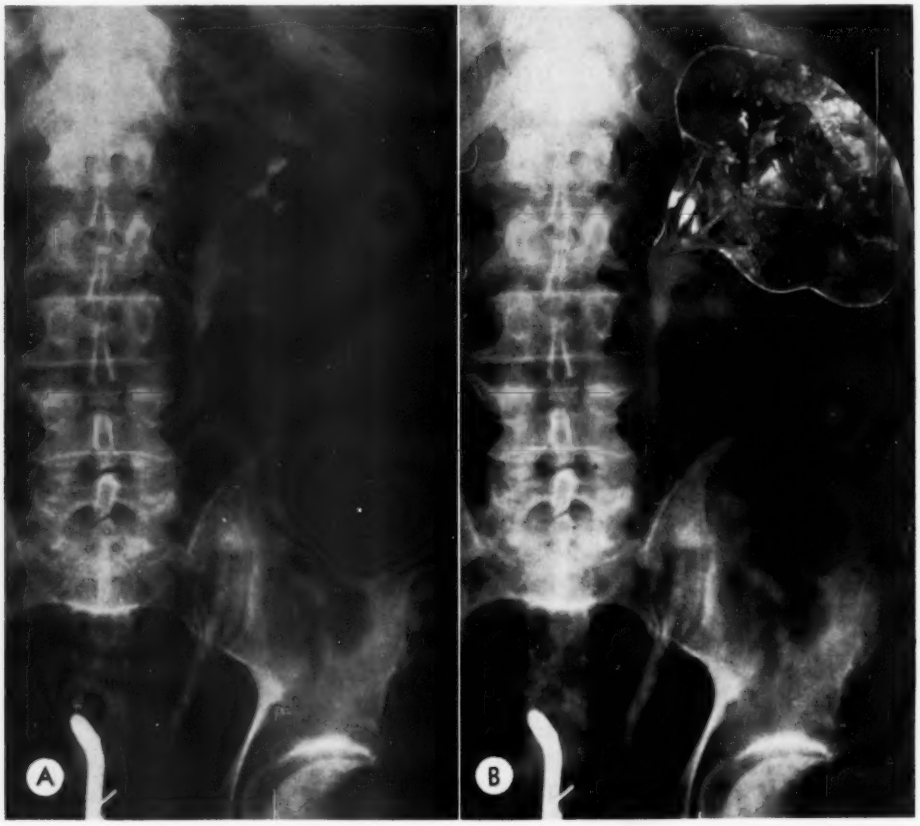

FIG. 4. Case IV. (*A*) Retrograde pyelogram showing deformity of the inferior calyces. (*B*) Composite photograph of the pyelogram and excised kidney.

decompensation secondary to a long history of rheumatic heart disease. Diabetes mellitus was present and there had been an episode of coronary thrombosis in 1955. Urinalyses showed albuminuria, mild pyuria, hyaline and granular casts. The patient expired and at autopsy a bilateral extrarenal calyceal system was found (Fig. 5). In addition, the left kidney exhibited malrotation and was located in an iliac ectopic position.

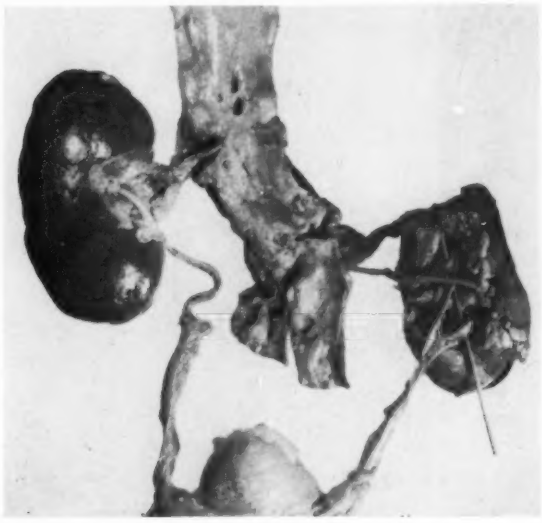

FIG. 5. Case V. Bilateral extrarenal calyces.

CASE VI. E.J. This sixty year old white male was admitted to the Veterans Administration Hospital with a history of gross hematuria of one year's duration. The patient admitted symptoms of prostatic obstruction. Physical examination revealed hypertension and residuals of a cerebral accident in 1957. The prostate was slightly enlarged at palpation and was benign. Urinalysis showed 15–20 red blood cells per high power field. An intravenous urogram and retrograde pyelogram demonstrated distortion of the left calyceal system, suggesting a space occupying lesion (Fig. 6*A*). At this time a diagnosis of extrarenal calyces was made. Surgical exploration disclosed two

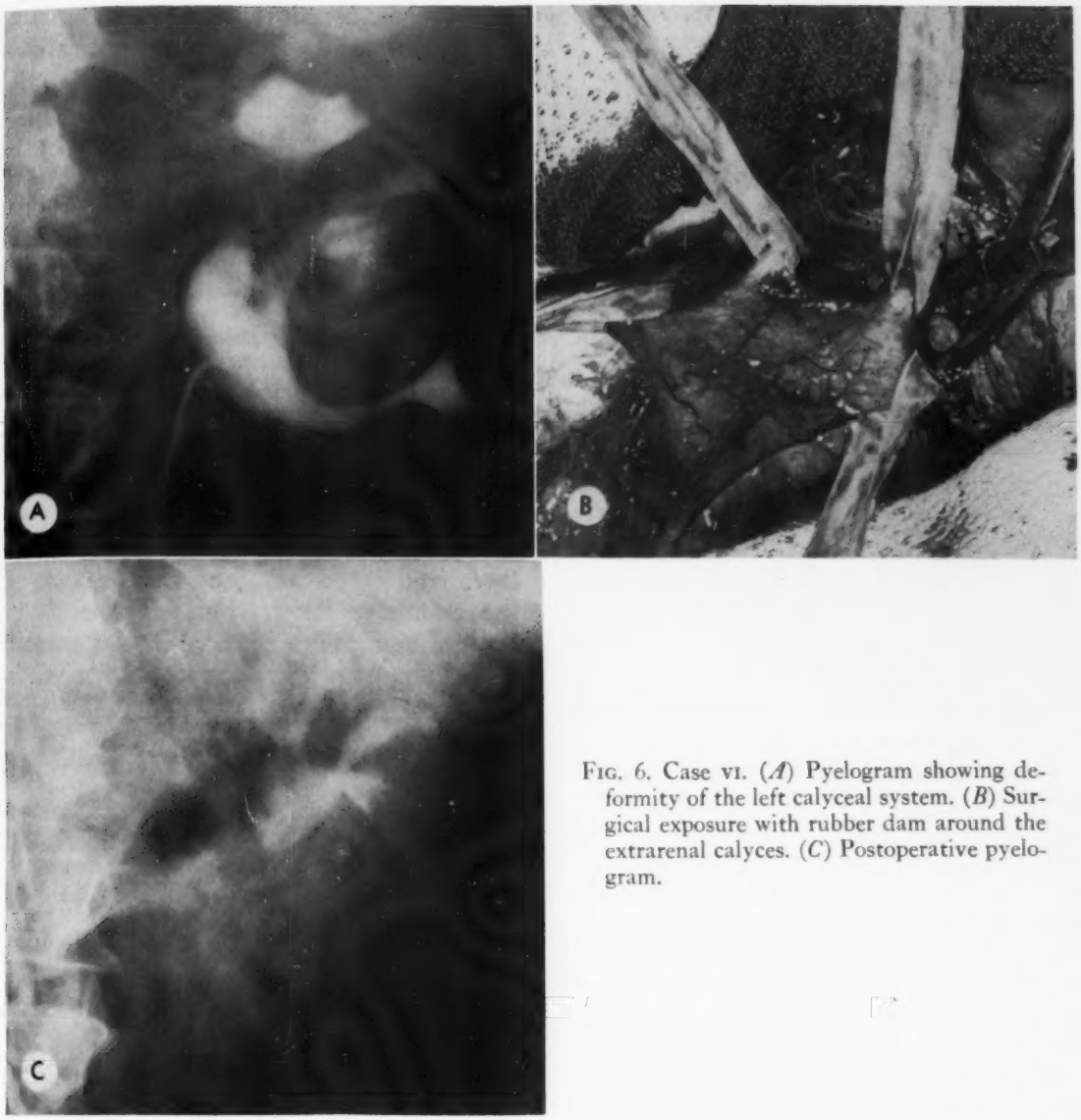

Fig. 6. Case vi. (*A*) Pyelogram showing deformity of the left calyceal system. (*B*) Surgical exposure with rubber dam around the extrarenal calyces. (*C*) Postoperative pyelogram.

large renal cysts and dissection of the renal hilus exposed the pelvis and four major calyces located outside the renal parenchyma, thus confirming the preoperative diagnosis of an extrarenal calyceal system (Fig. 6, *B* and *C*).

Case vii. H.S. This sixty-two year old white obese male was admitted to the Veterans Administration Hospital in 1960 with vague complaints of left flank pain. Urinary symptoms were minimal. An intravenous urogram and retrograde pyelogram revealed a large renal shadow with absence of the superior calyces, suggesting a possible superior pole tumor (Fig. 7). The pyelographic outline of the pelvis and calyces indicated an extrarenal position. Surgical exploration disclosed extensive perirenal lipomatosis and approximately 500 gm. of fat was removed. During the dissection the anticipated extrarenal calyces were identified.

Case viii. W.R. This forty-seven year old white male was admitted to the Veterans Administration Hospital in November, 1957, complaining of severe right flank pain radiating anteriorly and downward to the right lower quadrant. In 1952 a left pyeloplasty had been performed at another hospital. Because of persistent left flank pain, he had entered this hospital in 1953 at which time recurrent hydro-

FIG. 7. Case VII. Pyelogram showing a large tumor mass in the upper pole of the kidney, with extrarenal calyces.

nephrosis and a stricture of the ureteropelvic junction were found. A second pyeloplasty was performed with a good result. The patient had no complaints until the onset of the right flank pain three days prior to this admission. Pyelograms showed the presence of a right hydronephrosis due to ureteropelvic obstruction. Surgical exploration revealed extensive peripelvic adhesions and a taut anomalous blood vessel extending to the renal pelvis and kinking the ureteropelvic junction. Further exploration of the hilus demonstrated the presence of extrarenal calyces. No intrinsic stricture of the ureteropelvic junction was found and surgery was limited to resection of the redundant pelvis and division of the aberrant vessel.

COMMENTS

Fourteen cases of extrarenal calyces have been reviewed (Table 1), of which 8 cases occurred in our series. Of the total, 6 cases showed additional congenital abnormalities, namely, ectopia in 4 cases and partial ureteral reduplication in 2 cases. The left side was involved twice as frequently as the right. Renal disease was present in 8 cases and 7 of these could be related to the extrarenal calyceal deformity.

Statistics of renal disease associated with other renal congenital anomalies are not available. However, a kidney featuring a congenital deformity in size, shape, location or formation is known to exhibit a high incidence of renal lesions. The cause may be due to anomalous vessels, fibrous bands, and interference with urinary drainage due to its abnormal position or formation.

The diagnosis of a congenital anomaly is usually made when the clinical symptoms warrant a urologic investigation. In our series, except for the 2 cases found at autopsy, attention was focused on the extrarenal calyceal kidney not because of the anomaly, *per se*, but as a result of clinical symptoms induced by the renal pathology. However, the congenital mal-

FIG. 8. Case VIII. Appearance of the extrarenal pelvis of the kidney prior to the onset of obstructive symptoms.

TABLE I

CASES OF EXTRARENAL CALYCES

Reported by	Other Congenital Anomalies	Location	Renal Disease
Eisendrath[2]		Left	Hydronephrosis
Fürstner[2]	Bifid kidney with hypoplasia	Right	
Herz[2]		Bilateral	Right hydronephrosis
Apert[2]	Bifid kidney	Left	
Veseen[3]		Left	Hydronephrosis
Dunn and Gibson[1]	Ectopia	Left	Hydronephrosis
Malament *et al.*			
Case I A.C.	Ectopia	Right	
II J.M.		Left	Calculous hydronephrosis
III R.B.	Ectopia	Right	
IV J.L.		Left	Perirenal hematoma
V A.H.	Left ectopia	Bilateral	
VI E.J.		Left	Renal cysts
VII H.S.		Left	
VIII W.R.		Right	Hydronephrosis

formation could well have been the cause of the associated renal disease. A good illustration of this is Case VIII, where the patient had been followed with pyelographic studies for several years because of a left hydronephrosis. The right kidney offered little of interest (Fig. 8) and was neglected until the sudden onset of right flank pain revealed an acute hydronephrosis which, on exploration, was found to be due to kinking of the ureteropelvic junction of an extrarenal calyceal system. Consideration must be given to the fact that the multiple elongated infundibula of the extrarenal calyces, exhibiting peristaltic motion in a perirenal area, are more liable to extrinsic influence, as aberrant vessels, and may develop gradual or acute obstruction, as well as other complications.

SUMMARY

Extrarenal calyceal system is a congenital anomaly in which the renal pelvis and calyces lie outside the kidney and are attached to the renal parenchyma at the minor calyceal level.

The diagnosis may be made from routine pyelographic studies when the renal pelvis appears to be situated outside the renal shadow, and the calyces extend to the kidney with an incurvation resembling the prongs of a small hand garden rake.

Of the total of 14 cases reviewed, 8 occurred in our series, indicating that the incidence may be higher than previously suspected. As awareness of this congenital entity increases, more cases may be recognized.

Although extrarenal calyces may exist without causing any clinical symptoms, 50 per cent of the total series exhibited renal disease that could be attributed to this congenital malformation.

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HAMARTOLIPOMA OF THE KIDNEY: CLINICAL AND ROENTGEN FEATURES*

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BECAUSE of the presence of substantial amounts of fat, retroperitoneal tumors containing lipomatous elements can be recognized by roentgen examination. This was originally pointed out by Windholz in 1946.[22] The roentgen features indicating the presence of a retroperitoneal mass have been clearly described by several authors.[3, 4, 6, 9, 11, 23] The possibility of roentgenographically distinguishing a lipomatous tumor which arises within the kidney from similar tumors which arise from the retroperitoneal tissues has not been mentioned.

The present report summarizes the roentgen as well as the clinical features in 6 cases of lipomatous tumors arising within the kidney; these cases were collected at this hospital over a period of about nineteen years. It is evident from the small number of cases that this type of lipomatous tumor is uncommon. We believe, however, that it is of considerable clinical importance to separate lipomatous tumors arising within the kidney from other retroperitioneal tumors. This opinion is based, not only on the features of the current group of cases, but also on a review of the literature, which indicates that there is no documented case of an intrarenal lipomatous tumor which has metastasized.

REVIEW OF THE LITERATURE

In Robertson and Hand's[16] 2 personal cases and their summary of 12 cases from the literature, there was no example of a malignant lesion. In Klapproth, Poutasse and Hazard's[10] series of 4 personal cases and 12 analyzed from the literature, none were considered to be malignant. Williams and Savage[21] reported 1 personal case without metastases and reviewed 29 others from the literature. These latter authors considered all of these tumors to be "liposarcomas" of the kidney on the basis of histologic appearance and the presence of metastases in 2 cases, 1 reported by Tesler and Koiransky[19] and the other by Lubarsch as quoted by Pemberton and MacCaughan.[15] However, the case of Tesler and Koiransky[19] is clearly stated by the authors to be perirenal in origin and the case of Lubarsch is referred to by Pemberton and Mac Caughan[15] as also being perirenal.

Histologic criteria of malignancy based on the appearance of individual cells are uncertain. Vascular or lymphatic invasion has not been described. The fact that the capsule of the kidney may be penetrated by the tumor is not sufficient evidence of the malignant nature of these neoplasms. In 2 cases from this institution previously reported by Edelman[5] and brought up to date in this report (Cases 1 and 11), the designation "liposarcoma" was originally used on the basis of extension into the perirenal soft tissues. These patients have survived fourteen and nineteen years since nephrectomy without evidence of disease. Review of the histologic slides indicates no microscopic features to warrant the diagnosis of a sarcoma. In the series of 29 cases reviewed by Williams and Savage,[21] 11 patients were followed for more than one year without evidence of recurrence or metastasis. Survival for more than ten years was not uncommon in this series.

For practical purposes, therefore, it would appear that lipomatous tumors arising within the kidney are benign and that permanent cure may be anticipated if complete resection is feasible.

TERMINOLOGY

From the pathogenetic point of view, an

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unequivocal classification or designation of lipomatous tumors of the kidney is difficult and not generally agreed upon. In addition to substantial amounts of fat, these rumors usually contain smooth muscle elements and are often rich in blood vessels as well. As a result of their "mixed" character, such tumors have been variously designated as lipomata[16] or angiolipomata, fibrolipomata, angiofibrolipomata, etc.[2,10,12,13,17,18] Masses or tumors of exactly the same histologic and gross characteristics are not infrequently seen in patients with tuberous sclerosis[1,7,8,14] and in such instances have been referred to as hamartomata. According to Robertson and Hand,[16] careful search at necropsy indicates that small "lipomata" are found in about 1 per cent of all cases and are often multiple. Somewhat similar lesions within the liver have also been referred to as hamartomatas. On the basis that it is likely that lipomatous tumors of the kidney arise from congenital rests, the term "hamartolipoma" has been used in the title of this report. It should be emphasized that most patients with this tumor do not suffer from tuberous sclerosis. The designation hamartolipoma also serves to suggest the important clinical features of these tumors, namely, slow growth and benign nature. From the pure histologic point of view, however, the most suitable designation for these tumors is myolipoma or angiomyolipoma.

PATHOLOGY

In the early stages, a hamartolipoma of the kidney is entirely confined to the kidney parenchyma and is sharply demarcated from the normal kidney tissue. In places, a thin pseudocapsule may be evident but it is ordinarily not possible to enucleate the tumor cleanly from the kidney.

The fact that growth may be extremely slow is indicated by a case reported by Klapproth *et al.*[10] During a period of seventeen years, this patient complained of recurrent painless hematuria. When a lipomatous tumor was removed, it was only 1 cm. in diameter. Such tumors may, however, reach a huge size and one has been reported which attained a weight of 3,500 gm.[21] With growth of the tumor, there is usually compression and resultant deformity of the collecting system of the kidney. Rarely, if ever, is there direct infiltration into the pelvis or the calyces. More commonly, growth of the tumor is peripheral, into and through the renal capsule, but even in such instances the adherent and infiltrated capsule can ordinarily be identified.

In many cases, satellite nodules of similar character may occur within the kidney substance at some distance from the main tumor. The presence of more than one tumor within the kidney has sometimes been interpreted as evidence of local metastases[12,21] but it seems equally likely that a multicentric origin is the cause of this finding.

CLINICAL FEATURES

In the absence of tuberous sclerosis, hamartolipomas of the kidney are most commonly evident in the third to the fifth decades. It is frequent to obtain a history of a mass being present and growing slowly for several years prior to the time of surgical intervention. According to the literature, in the majority of instances, these tumors are detected as incidental findings because of the presence of a mass on abdominal palpation. In other cases, the patient presents with a history of an episode or episodes of rather acute flank or abdominal pain. This was true in 5 of the 6 cases included in this report. In such cases, operative intervention has demonstrated hemorrhage within or around the tumor or kidney which presumably was the cause of the pain. Hemorrhage or hematoma formation of this type is usually seen in the absence of any history of significant trauma.

The fact that hemorrhage is common is of importance to the radiologist because the characteristic roentgen feature of large lucent areas within a tumor mass may be partly obscured by the blood. Moreover,

perinephric hematoma formation may show all the roentgen features of a perinephric abscess. Hematuria is not a common finding. It was present in only 1 patient in the 6 cases described in this report.

In all reported series, the incidence in females is greater than in males by 2 or 3 to 1. Of the 6 cases included in this report, all were females.

These tumors are bilateral in a significant number of cases, perhaps about 10 per cent.[21] In 2 of the 6 cases described below, the tumors were bilateral. This feature is of great clinical importance and must be carefully investigated in every case, since these tumors are difficult to eradicate without nephrectomy. If further experience should confirm the opinion that these tumors do not metastasize, it would appear to be reasonable not to risk the loss of normal renal tissue by attempting operative intervention on either side when bilateral tumors are present. In such cases, it is likely that both kidneys will eventually be completely destroyed and the patient will die of uremia, but this is not likely to occur for many years.

REPORT OF CASES

CASE I. I.E. This was a forty-one year old female admitted in 1946 with the chief complaint of sharp intermittent pain in the left lower quadrant of two days' duration. The pain was accompanied by nausea and vomiting and radiated toward the back. There was a history of low back pain for a period of about thirteen years.

Physical examination was negative except for the presence of a large tender mass in the left flank and moderate costovertebral angle tenderness. Hemoglobin was 75 per cent, white blood cell count 8,600 cells per ml., with a normal differential count. Urine examination was not remarkable. Blood urea was not elevated. Intravenous pyelography was reported as showing distortion of the calyces in the upper pole of the left kidney. This was confirmed by retrograde pyelography. The appearance was described as characteristic of a renal neoplasm. A left nephrectomy was performed a few days after admission with the preoperative diagnosis of hypernephroma.

The specimen consisted of the kidney and the investing Gerota's fascia. A considerable amount of blood was present in the adherent perirenal soft tissues. The kidney weighed 450 gm. and appeared to be enlarged and markedly deformed. The lower two-thirds of the kidney was replaced by a sharply circumscribed encapsulated tumor about 12 cm. in length. The capsule of the kidney could easily be stripped except at the area of bulging of the tumor. On section, a markedly yellow tumor was found which surrounded and distorted the lower portion of the pelvis and the lower calyces; the upper third of the kidney appeared normal.

Histologic diagnosis was originally a leiomyoliposarcoma. The patient is alive and well fourteen years after nephrectomy. The pathologist has reviewed the slides and has changed the diagnosis to a benign tumor.

Comment: This patient is entirely well fourteen years after nephrectomy for an intrarenal lipomatous tumor. The tumor was discovered because of an acute episode of pain, although "low back pain" had been present for many years. A perirenal hematoma was found at exploration. The original pyelograms of this case are not available for review. It is likely, however, in view of the operative findings, that lucent areas within the tumor might have been evident on careful search.

CASE II. S.M. This was a forty-six year old female admitted to this hospital in 1941 for exploration of a left flank mass known to have been present for at least two years. It had been discovered during examination after an automobile accident. There had been no evident increase in the size of the mass during the two year period of observation. Intravenous pyelography performed one year prior to admission demonstrated that the mass was in the left kidney and had compressed and distorted the inferior and middle calyces.

Physical examination at the time of admission was negative except for a grapefruit-sized mass in the left flank, which was ballotable but not tender. A plain roentgenogram of the abdomen did not show a distinct left renal outline (Fig. 1*a*). Intravenous pyelography again demonstrated compression and elevation of the middle infundibulum as well as the lower portion of the pelvis of the kidney (Fig. 1*b*). A

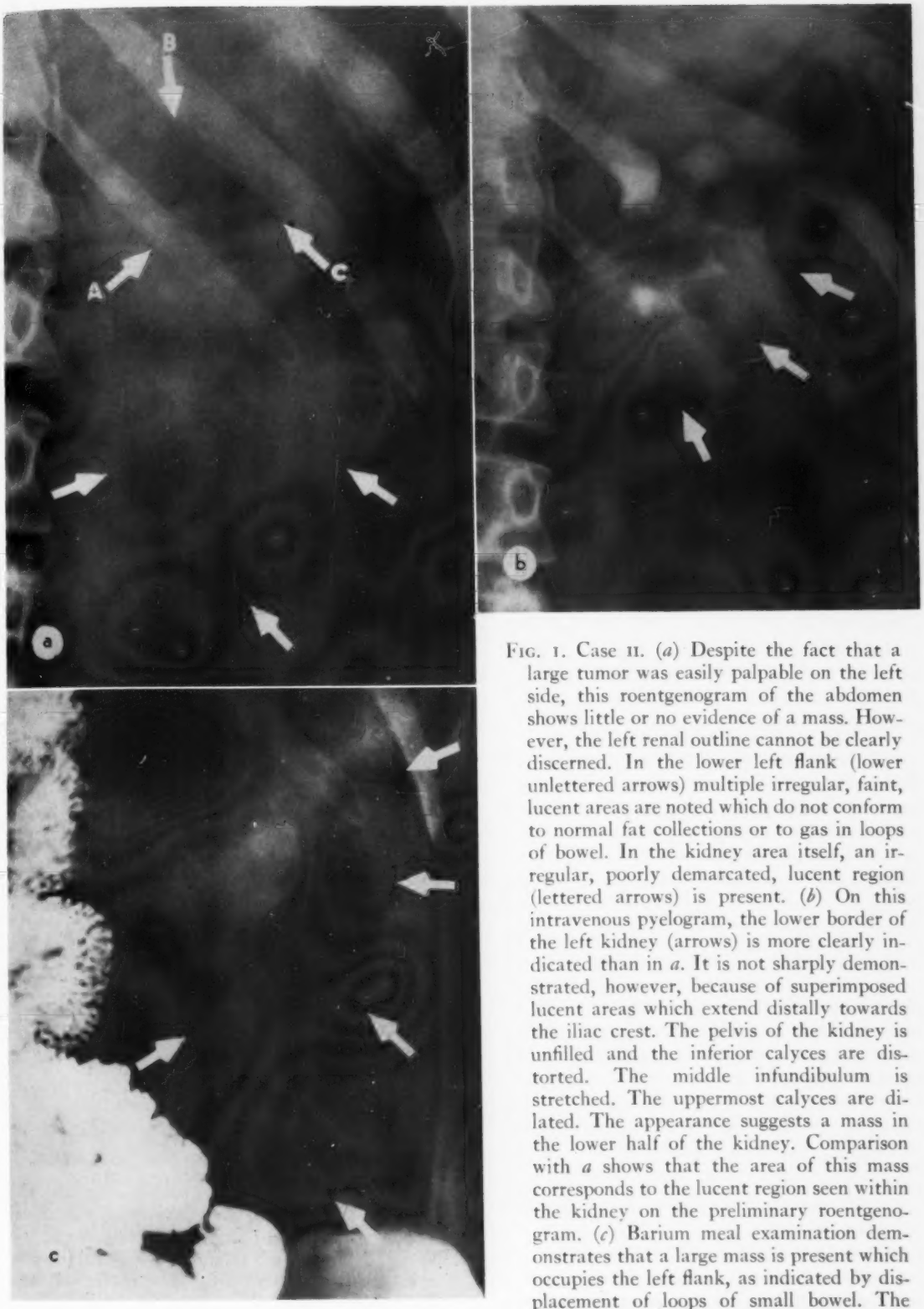

Fig. 1. Case II. (*a*) Despite the fact that a large tumor was easily palpable on the left side, this roentgenogram of the abdomen shows little or no evidence of a mass. However, the left renal outline cannot be clearly discerned. In the lower left flank (lower unlettered arrows) multiple irregular, faint, lucent areas are noted which do not conform to normal fat collections or to gas in loops of bowel. In the kidney area itself, an irregular, poorly demarcated, lucent region (lettered arrows) is present. (*b*) On this intravenous pyelogram, the lower border of the left kidney (arrows) is more clearly indicated than in *a*. It is not sharply demonstrated, however, because of superimposed lucent areas which extend distally towards the iliac crest. The pelvis of the kidney is unfilled and the inferior calyces are distorted. The middle infundibulum is stretched. The uppermost calyces are dilated. The appearance suggests a mass in the lower half of the kidney. Comparison with *a* shows that the area of this mass corresponds to the lucent region seen within the kidney on the preliminary roentgenogram. (*c*) Barium meal examination demonstrates that a large mass is present which occupies the left flank, as indicated by displacement of loops of small bowel. The numerous partially confluent lucent areas (arrows) are more clearly shown in this reproduction. These extend into the kidney substance.

barium meal examination (Fig. 1*c*) showed marked displacement of the jejunal loops toward the midline. A left nephrectomy was performed.

In the posterior aspect of the kidney were several masses, the largest of which measured 12 cm. in diameter. These masses appeared to be well demarcated from the kidney parenchyma and the portions that extended outward from the kidney were clearly delineated by a thin-walled translucent, in places hemorrhagic, capsule. On cut section, it was found to be soft and uniformly golden-yellow except for a large 3.5×3.5 cm. area near the kidney surface which was grayish-yellow and firmer in consistency. The pelvis of the kidney was compressed by the mass. A large portion of normal kidney was preserved.

The histologic diagnosis was lipomyosarcoma of the kidney but the pathologist included in his report the observation that the diagnosis of sarcoma was based on what was presumed to be an infiltrating character of the tumor as described by the surgeon.

Comment: In this patient, an asymptomatic mass in the left kidney was discovered incidentally and did not change in size in a period of two years. This patient is alive and well nineteen years after nephrectomy. Review of the original roentgenograms shows lipomatous, *i.e.*, lucent areas, within the confines of the kidney tumor (Fig. 1, *a*, *b* and *c*).

CASE III. B.S. This twenty-four year old female was admitted with the chief complaint of a painful mass in the right flank. This mass, noticed by the patient herself six months prior to admission, was said to have increased recently in size. Originally, the mass was not painful or tender. However, seven days prior to admission, she began to complain of pain on the right side of the abdomen which radiated to the back. The pain increased in severity and was accompanied by nausea and rigidity of the right side of the abdomen. Within a period of four hours, however, the pain diminished. She was hospitalized elsewhere and it is known that the urine examination at that time showed no evidence of blood. Five days prior to admission to this hospital, she showed temperature increases to 101° F. She received a blood transfusion one day prior to admission.

On admission, physical examination was negative except for a grapefruit-sized mass in the right upper and lower quadrants. The mass was smooth and not tender. Laboratory examination showed a hemoglobin of 13.5 gm. per 100 cc., a white blood cell count of 4,500 cells per ml. with a shift to the left. The urine was negative. Cystoscopy was within normal limits. Barium enema examination and gallbladder study were negative except for marked caudad displacement of the hepatic flexure. Retrograde pyelography (Fig. 2) revealed the collecting system of the right kidney to be displaced towards the midline. In the kidney and immediately adjacent to the pelvis, multiple partially confluent lucent areas were evident. The right psoas margin could not be identified. There was a minimal scoliosis of the lumbar spine with convexity to the left. The presence of the lucent areas replacing the kidney parenchyma led to the preoperative

FIG. 2. Case III. Right retrograde pyelogram. The renal outline cannot be discerned. The renal shadow is partially occupied by irregular lucent areas (arrows) which extend to the collecting system. The calyces are blunted or flattened. The collecting system and ureter are displaced towards the midline. The psoas margin is obliterated.

diagnosis of a lipomatous tumor of the kidney. Laparotomy revealed a large mass arising from the right kidney and displacing the hepatic flexure downward. A smaller mass of similar consistency was felt in the left kidney. A right nephrectomy was performed.

The specimen showed a large subcapsular hemorrhage in the upper pole. Under the hemorrhage, there was a mass about 7 cm. in diameter in the substance of the kidney. It was yellow in color with several grayish-pink, firm areas within it. A number of yellowish satellite masses were scattered throughout the remaining portion of the kidney. The renal pelvis appeared normal.

The histologic diagnosis was multiple myolipomata of the kidney.

Comment: At operation in this patient, there was evidence on palpation of a soft, small mass in the left kidney in addition to the large tumor of the right kidney. This is presumed also to be lipomatous. The roentgenogram of the abdomen taken during right retrograde pyelography shows no definite lucent areas within the left kidney outline (Fig. 2). The patient's history of recent pain and increase in size of the mass may be attributed to the subcapsular hematoma found at operation. This patient is alive and well one year after discharge.

Case iv. L.L. This fifty-one year old female was admitted with the chief complaint of right flank pain of five days' duration. The history revealed intermittent episodes of left renal colic for the preceding six years.

Physical examination was negative except for a tender mass in the right flank. Laboratory examination revealed a hemoglobin of 9.6 gm. per 100 cc. and a white blood cell count of 7,550 cells per ml. with a normal differential count. The urine analysis was normal. Sedimentation time was 100 mm. per hour. A plain roentgenogram of the abdomen showed obliteration of the right psoas outline and a moderate scoliosis of the lumbar spine. The kidney outline was obscured but appeared to be considerably enlarged. Intravenous pyelography revealed a moderate medial displacement of the collecting system of the right kidney and marked deformity and compression of the superior calyx and infundibulum (Fig. 3*a*). The diagnosis of a perinephric collection, presumably blood because of anemia, was made. At operation, a tense blueish mass was found in the right flank near the upper pole of the kidney. Nephectomy was performed.

The specimen showed a mass 5×5 cm. within the upper pole of the kidney with areas of fat and hemorrhage. There was a large subcapsular hematoma. The lower two-thirds of the kidney was normal.

Histologic diagnosis was myolipoma with subcapsular hematoma. This patient died two years after nephrectomy from a coronary occlusion. Autopsy was not obtained. There had been no complaints referable to the urinary tracts since the time of operation.

Comment: Careful retrospective examination of the intravenous pyelogram of this patient shows a faint lucent area adjacent to the deformed superior infundibulum and calyx (Fig. 3*a*). This was somewhat better seen on a roentgenogram of the abdomen taken six years previously (Fig. 3*b*) during an episode of left renal colic due to a small calculus. It is likely that the hemorrhages within the tumor as well as the subcapsular hematoma served to obscure the fatty areas.

Case v. G.S. This twenty-nine year old female was admitted with the chief complaint of right flank pain of three weeks' duration. The pain started suddenly with radiation to the right shoulder while the patient was working in the office and necessitated morphine injection. There was no hematuria or previous significant history. Two weeks prior to the onset of the severe pain, because of vague abdominal symptoms, she had had a gallbladder series which was said to have shown stones.

Physical examination on admission was negative except for a large mass in the right flank. Hemoglobin was 9.4 gm. per 100 cc. White blood cell count was 12,700 per ml. with a shift to the left. Urine was negative. Intravenous pyelography (Fig. 4*a*) revealed somewhat diminished function on the right. The pelvis and calyces were stretched and appeared to be uniformly narrowed and displaced toward the midline. The renal outline could not be discerned. In retrospect, numerous lucent areas can be seen within the kidney area and extending inferiorly towards the iliac crest. The most marked deformity of the collecting system was

FIG. 3. Case IV. (*a*) Intravenous pyelogram shows displacement of the right renal pelvis towards the midline. The superior infundibulum and calyx (arrow) are narrowed and deformed. The renal outline and psoas margin are obscured. On the original roentgenogram, a faint lucent area can be discerned adjacent to the deformed superior calyx. At operation, an intrarenal lipoma with numerous hemorrhagic areas was found in addition to a large subcapsular hematoma. (*b*) Roentgenogram taken six years before *a* because of left renal colic due to a ureteral stone. A faint lucent zone (between arrows) about 1½ inches in diameter can be discerned within the upper pole of the right kidney. The outline of the kidney is clearly evident at this time.

an inferior prolongation and stretching of the lowermost infundibulum. The significance of these roentgen findings or the significance of somewhat similar lucent areas within the outline of the left kidney was not appreciated preoperatively. After preparation with multiple transfusions, the patient was explored and a right nephrectomy was performed.

The specimen consisted of a kidney weighing 1,375 gm. and measuring 23×9.5×5 cm. The surface of the kidney was lobulated but the reniform shape was retained. The capsule was stripped with difficulty and revealed a grayish-red surface showing numerous yellowish nodules varying in size from 3 mm. to 9 cm. The largest of these masses occupied the lower pole of the kidney. A section of the kidney revealed it to consist of numerous yellowish, glistening, fleshy nodules and hemorrhagic cystic areas filled with blood clot. There were only small areas of residual renal parenchyma; within these areas tiny yellow nodules were also noted. There was no evidence of invasion of the ureter, renal pelvis or major branches of the renal artery and veins.

Histologically, the tumor was a myolipoma with no evidence of malignancy.

As a result of the findings, the pyelograms were reviewed and attention was called to the lucencies in the left kidney. Careful palpation of the left renal area confirmed the presence of a mass. Three months after the first operation, the patient was readmitted and intravenous pyelography was repeated. There was dilatation of the pelvis and calyces of the left kidney (Fig. 4*b*), which, however, did not appear to be remarkably distorted or compressed. Only

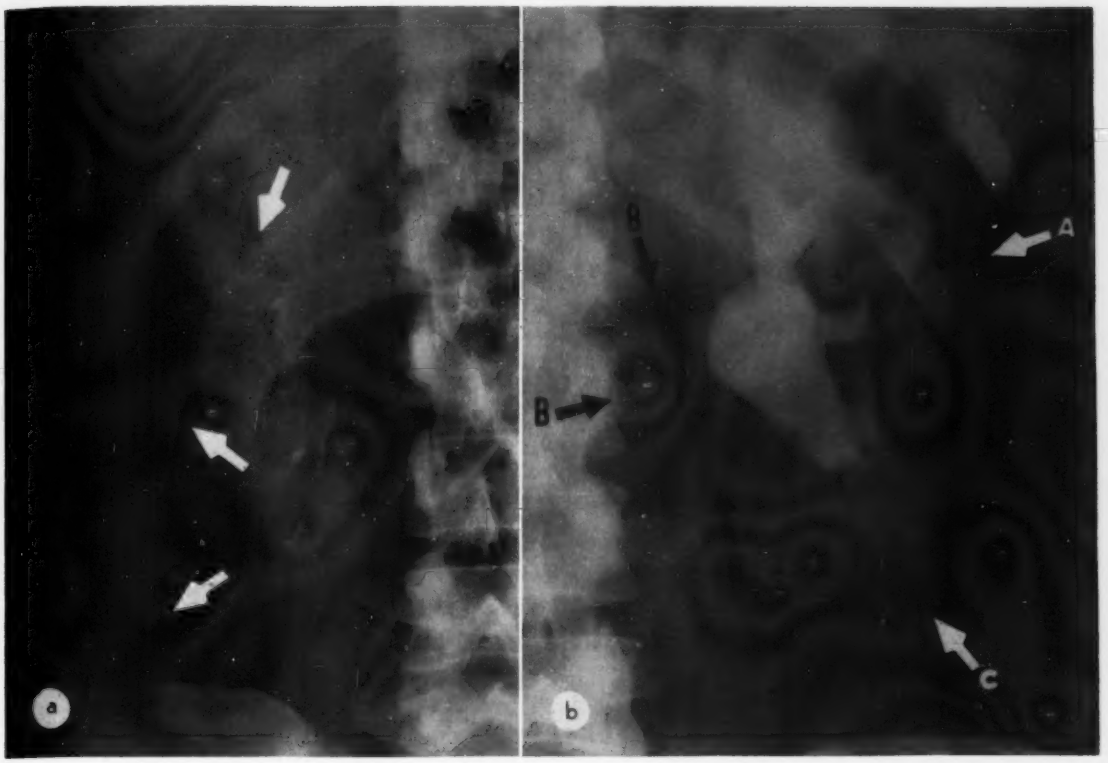

FIG. 4. Case V (*a*) Intravenous pyelogram shows compression of the entire collecting system of the right kidney. The kidney outline is absent. Numerous band-like and more globular lucent areas (arrows) with intervening areas of water density occupy the renal area and extend inferiorly. (*b*) Intravenous pyelogram three months after right nephrectomy shows moderate dilatation of the left pelvis and calyces. Only the upper lateral portion of the kidney outline (arrow A) is clearly demarcated from adjacent normal perirenal fat. Within the lower two-thirds of the kidney, there are numerous discrete and confluent lucent areas which replace the normal renal outline and extend inferiorly toward the iliac crest (arrow A to arrow C). Medially, adjacent to the pelvis (arrows B), there is a more discrete globular lucent area which extends to the spine.

the upper portion of the lateral part of the left renal outline could be identified. The lower pole was completely obliterated or effaced by an irregular, mottled, poorly demarcated group of shadows with many lucent areas within it. Apparently located medial to the pelvis of the kidney there was a somewhat more discrete, lucent, ovoid area with streaks of water density within it. The left renal area was explored and a mass was found within the kidney extending into the adjacent soft tissues. It was clear, however, that this tumor could not be removed without removing the kidney. As a result, only a biopsy was performed. The biopsy was reported as myolipoma. Postoperatively, the patient bled briskly from the wound. This was brought under control, but ten days after discharge from the hospital rather massive bleeding recurred and the patient died.

Comment: This patient demonstrates the difficult clinical problem when renal lipomata are bilateral. In retrospect, it would have been wiser not to have explored the second kidney to determine the possibility of removal. In fact, it can be argued that, if the status had been correctly evaluated, neither kidney should have been explored.

CASE VI. L.G. This was a fifty-two year old female who entered the hospital with the chief complaint of pain in the left flank. There was no history of hematuria.

Physical examination was negative except for a ballotable and slightly tender mass in the left flank. Blood pressure was 130/70. Laboratory examination showed a hemoglobin of 9.6 gm., a white blood cell count of 12,400 per ml. with a slight shift to the left. Urine examina-

FIG. 5. Case VI. Intravenous pyelogram shows elevation of the collecting system and stretching of the inferior calyces. Only the superior lateral margin of the kidney outline (upper arrow) is evident. Within the lower pole of the kidney, a globular lucent zone (lower arrows) is present which is capped inferiorly by a large crescentic area of water density. The psoas margin can be faintly seen through the mass.

tion was not remarkable. Sedimentation time was 4 mm. in the first hour. Roentgenography of the abdomen and intravenous pyelography (Fig. 5) showed a mass in the lower pole of the left kidney. There was displacement and distortion of the lowermost calyces. The renal outline of the lower half of the kidney could not be seen. Within the lower pole of the kidney, there was a very poorly demarcated, markedly lucent, somewhat spherical lesion with faint, somewhat beaded, linear densities extending through it. At operation, the presence of a mass in the lower pole of the left kidney was confirmed and nephrectomy was performed.

The specimen showed a rather soft mass about 12 cm. in diameter. It was well circumscribed, was entirely within the kidney parenchyma and contained areas of necrosis and hemorrhage. In discrete areas, distinct yellowish fat was discernible.

Histologic report was myolipoma of the kidney. There was no evidence of any malignant change. The patient is alive and well four years after nephrectomy.

Comment: The roentgen features (Fig. 5) in this case correspond exactly to the anatomic findings, since this lesion was discrete and was confined to the kidney.

ROENTGEN FEATURES

The roentgen features of lipomata of the kidney may be divided into two groups: (1) findings indicative of a mass and (2) findings indicative of fatty tissue within the mass. The first group includes deformity and compression of the kidney pelvis and calyces characteristic of any intrarenal mass (Fig. 1*b*). In the case of a lipoma, invasion of the collecting system and bizarre deformities are unlikely. Displacement of the pelvis and calyces as a whole (Fig. 2 and 5) is a more common finding in a lipomatous tumor than in a hypernephroma, presumably because of its slow growth. This generalized displacement of the collecting system may be quite marked in the larger tumors when extrinsic growth is extensive. In such cases, there may also be obvious displacement of adjacent small bowel loops (Fig. 1*c*) and colon.

The presence of a renal lipomatous tumor which, clinically, is easily palpable may be completely overlooked roentgenologically unless pyelography is performed. Moreover, if attention is given exclusively to the deformed collecting system, the extent of the tumor will usually be underestimated. The reason for this is that the usual roentgen feature of a large kidney mass, that is, a uniformly dense globular area continuous with but protruding from the kidney outline, is absent. Since a lipomatous tumor has a decreased density, the kidney outline is completely lost or markedly obscured at the site of the tumor (Fig. 1*b*, 2, 4*a*, 4*b*, and 5). Moreover, no clear-cut periphery of soft tissue or water density is present outlining the limits of the tumor. Instead, a nondemarcated, nondescript, mottled shadow consisting of irregular lucent zones interspersed with

Fig. 6. Perirenal lipoma. The sharply outlined lateral aspect of the kidney is compressed and straightened (medial arrows). The kidney is elongated and displaced toward the midline. The pelvis and calyces are within normal limits. A mass composed of mottled lucent areas is immediately adjacent to the kidney. The lower margin of the tumor is demarcated by a capsule of water density (lower two arrows) from the adjacent normally homogeneous retroperitoneal fat.

equally irregular areas of water density is present (Fig. 1*c*, 2, and 4, *a* and *b*). The lucent areas may be quite obvious (Fig. 2 and 5) but frequently they are so faint that intentional and careful search for them must be made. It may be necessary to repeat the roentgenographic study with better technique before the appearance is convincing. In most instances, the configuration of the lucent areas has no resemblance to gas in the bowel. Moreover, a gas filled portion of the bowel shows considerably greater lucency than a collection of fat of similar size. If there is any question, repeat examination including oblique views will demonstrate the constancy of the lucent areas and confirm their relationship to the kidney or retroperitoneal area. Theoretically, laminagraphy should also be useful for this purpose. In those cases in which the lucent areas are immediately adjacent to the pelvis and calyces (Fig. 2 and 5), there can be little doubt of the intrarenal location of the tumor. Unfortunately, even in such instances, hemorrhage within the tumor may obscure the original lucent zone (Fig. 3, *a* and *b*). Further diagnostic difficulty may arise as a result of perirenal hemorrhage which may exactly simulate, roentgenologically, a perinephric abscess.

In contrast to an intrarenal lipomatous tumor, a retroperitoneal tumor containing fat rarely shows evidence of extension into the kidney substance or deformity of the pelvis or calyces indicative of an intrinsic mass. There may be extrinsic compression of the kidney or marked dislocation of this organ as a whole. A distinction can sometimes be made between a perirenal lipomatous tumor (Fig. 6) and a similar tumor arising elesewhere in the retroperitoneal tissues on the basis of the location of the mass. Perirenal lipomatous tumors may have a somewhat better prognosis than other retroperitoneal tumors because complete excision is more likely to be feasible.

Renal lipomata must also be distinguished from renal replacement lipomatosis. This condition is usually associated with inflammatory disease and contraction of the kidney such as occurs in calculous pyelonephritis. Excess inflamed fat is found within the hilus of the kidney as well as surrounding the kidney. The outer aspects of the opacified pelvis and calyces may be rather uniformly outlined by such fat deposits. Recognition of the more uniform distribution of this fat and the basic underlying disease serve to differentiate this condition from a lipomatous tumor.

Since lipomata of the kidney may be multiple and bilateral, the deformity of the pelvis and calyces may simulate polycystic kidneys.[10] It is essential in such cases to recognize the numerous lucent zones indicative of the presence of fat within the masses.

There are 4 cases of hamartolipomata of the kidney reported in the literature[10,13,20,21] in which aortography was utilized for diagnostic purposes. Puddling of the opaque material as well as vascular displacements were demonstrated which resembled the findings seen with hypernephromata. It would seem, therefore, that this is not a useful technique for differential diagnosis in such instances.

SUMMARY

Lipomatous tumors of the kidney are uncommon lesions. Their true incidence is difficult to establish because of confusion in terminology. Such tumors have been variously designated as lipomata, liposarcomata, angiolipomata, leiomyolipomata, angiofibrolipomata nephrolipomata, and hamartomata. Histologically, the term "myolipoma"seems most apt, since muscle elements as well as fat are present. Pathogenetically, the term hamartolipoma appears most suitable, since it is likely that such tumors arise from embryonic rests.

A review of the literature as well as the features of the reported group of cases indicates that lipomatous tumors of the kidney are benign. There is no documented case which has shown evidence of metastases. For this reason, it is of clinical importance to distinguish this type of tumor from similar or mixed retroperitoneal tumors, which metastasize in a significant number of cases. Moreover, complete surgical removal of an intrarenal lipoma is usually less difficult than removal of a retroperitoneal tumor.

Six cases of renal hamartolipomata are presented in this report. In 5, the presenting symptom was a painful mass. All cases were in females. None suffered from tuberous sclerosis. In 2 cases, bilateral tumors were present. All patients were operated upon. Four are alive and well nineteen, fourteen, four and one year after surgery. One patient died subsequent to exploration of the left kidney after right nephrectomy, and the sixth from coronary occlusion.

The roentgen features of renal lipomata are usually sufficiently clear-cut to warrant a correct preoperative diagnosis. The diagnosis is based on the demonstration of a mass within the kidney containing lucent, *i.e.*, fatty, areas.

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PULMONARY HAMARTOMA*

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ALTHOUGH, according to Bateson and Abbott,[1] only 253 cases of pulmonary hamartoma were on record up to October, 1960, the literature is now so comprehensive that it seems superfluous to survey the history and the gross, microscopic, and usual roentgen findings of the lesion.

The object of the present paper is to report an additional 10 cases which differ in various ways from the series published so far.

All the patients were operated upon in the Departments of Thoracic Surgery of the University Hospital, Copenhagen, during the period 1951–1960, and the diagnosis was confirmed histologically in each case.

REPORT OF CASES

CASE I (Fig. 1, *A* and *B*). A female, aged five, with a history of operation for hiatal hernia at the age of three. After the operation, she thrived but continued having attacks of vomiting. Follow-up roentgen examination of the esophagus two years later showed a rounded well-defined, large mass located basally in the left lung, behind the heart, and unrelated to the esophagus. There was no visible calcification within the contours of the lesion. Left sided thoracotomy was performed. Posteriorly in the lower lobe, a firm, nodular tumor the size of a tangerine was palpable. Lobectomy and herniotomy were done and the postoperative course was uneventful. Gross and microscopic study revealed a typical hamartoma.

CASE II (Fig. 2). A male, aged fifty-six, who had always been in good health. At a routine examination, the chest roentgenogram showed a cherry-sized, homogeneous, smooth-margined, well-defined, nonlobulated opacity situated anteriorly in the right upper lobe. There was no visible calcification or cavitation. The surrounding pulmonary tissue was normal. Laboratory studies and bronchoscopy showed no abnormalities. Right-sided thoracotomy revealed, anteriorly in the upper lobe and close to the surface, a round, firm nodule which was shelled out easily. Gross and microscopic examination showed a typical hamartoma. The postoperative course was uneventful.

CASE III (Fig. 3, *A* and *B*). A female, aged ten, who had previously been in good health. Three months before admission, she began to have a cough productive of blood-tinged sputum. Laboratory findings were normal; in particular, there were no signs of tuberculosis. Chest roentgenography revealed atelectasis of the apical segment of the left upper lobe. Laminagraphy disclosed one large and several smaller cavities within the infiltrated area, but no visible calcification. On left-sided thoracotomy, the apicoposterior segment of the upper lobe was found to be atelectatic, shrunken and firmly infiltrated. The area was resected and the postoperative course was uneventful. Gross and microscopic study revealed a typical hamartoma with a walnut-sized cavity lined with respiratory epithelium.

CASE IV (Fig. 4). A male, aged thirty-eight, who had been in good health up to the age of twenty. After that age, he had had repeated episodes of hemoptysis, but there had been a symptom free period of about thirteen years. During the two years preceding hospitalization, he had again developed hemoptyses and cough. Laboratory studies showed no abnormalities; specifically, no tubercle bacilli. Bronchoscopy revealed some sanguineous secretion in the branch leading to the apical segment of the right lower lobe, but no other abnormalities. Roentgenograms of the lungs showed an ill-defined infiltration, the size of the palm of a hand, at the site of the right lower lobe, but no cavitation or calcification. The same appearances were seen at laminagraphy, and bronchography showed normal conditions apart from some shrinkage of the lower lobe. Right-sided thoracotomy was performed with removal of the lower lobe, which was small and partially atelectatic. No tumors were palpable but, after the bronchial branches had been cut,

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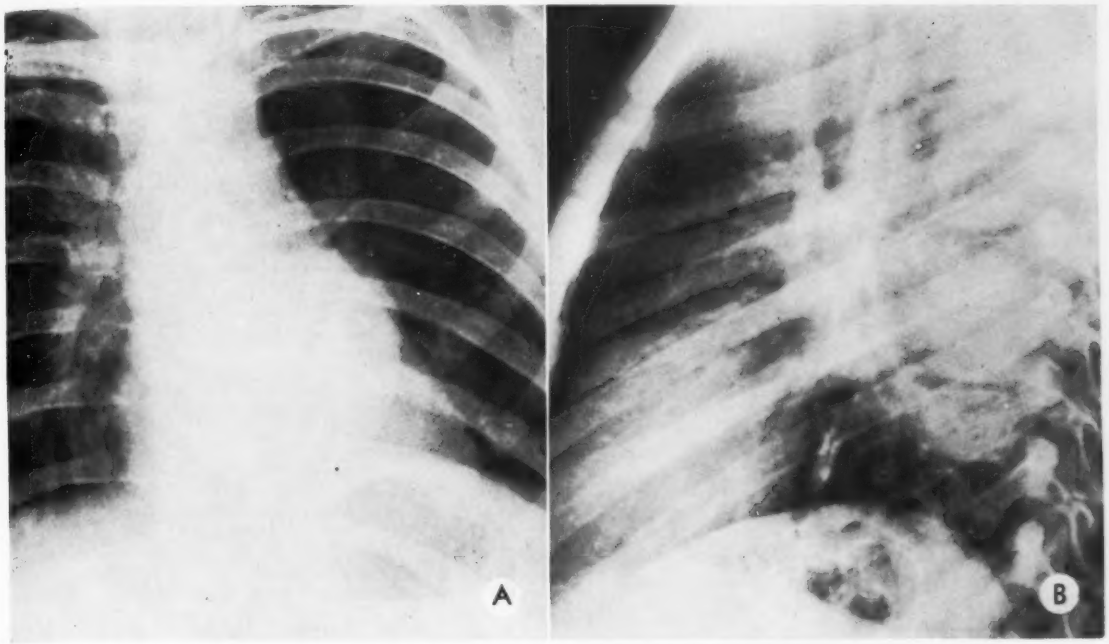

Fig. 1. Case I. (*A*) Big, round, well-defined hamartoma in the left lower lobe, visible through the heart shadow. (*B*) Lateral view.

the mucous membrane was found to be greatly thickened and vascular immediately before the branching of the bronchus to the apical segment. Histologic examination of this area revealed a typical hamartoma. The postoperative course was uneventful.

Case V (Fig. 5, *A* and *B*). A male, aged sixty-eight, who had always been in good health. Six weeks prior to admission, he started having cough and hemoptyses, but no other symptoms. Laboratory findings were normal. Bronchoscopy showed a grayish-red tumor in the bronchus to the apical segment in the right lower lobe. Roentgenograms of the lungs revealed a large, somewhat rounded, and fairly well-defined opacity beneath and behind the right hilus, at the site of the apical segment of the lower lobe. On the anteroposterior views it was partially hidden by the heart. There was no cavitation or calcification within the opacity. Bronchography showed nonfilling of the apical segment of the lower lobe, but no other abnormalities. Right-sided thoracotomy disclosed grape-like, firm, whitish masses of tumor tissue in the apical segment of the lower lobe, and the segment was resected. There was an uneventful postoperative course. Histologic examination showed a hamartoma.

Case VI (Fig. 6). A male, aged forty, with a history of right-sided pleurisy during adolescence. No other serious illnesses had been experienced; specifically, none with pulmonary symptoms. While he was in the hospital be-

Fig. 2. Case II. Small hamartoma in the anterior segment of the right upper lobe.

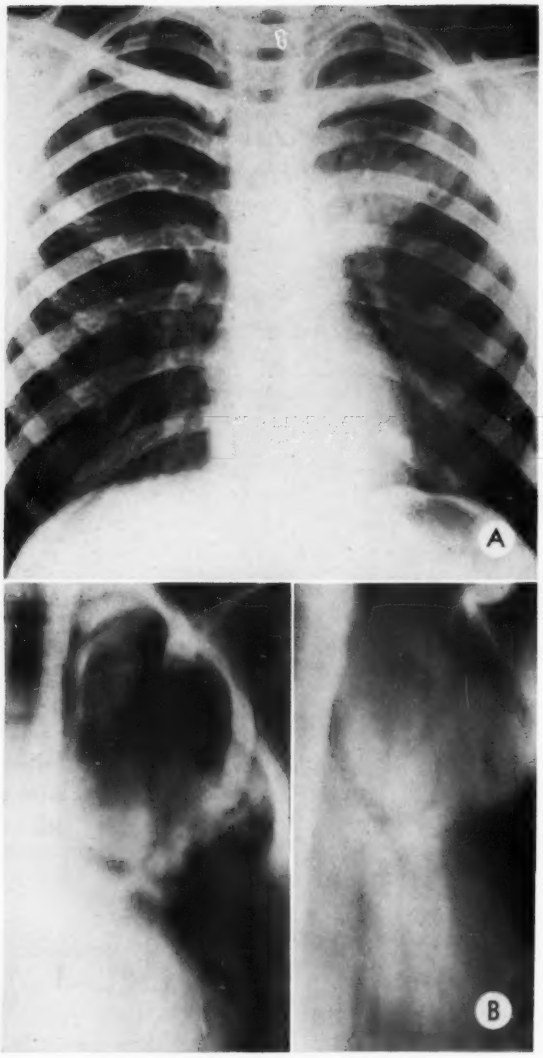

FIG. 3. Case III. (*A*) Segmental collapse of the left upper lobe due to an endobronchial hamartoma. (*B*) Laminagrams showing translucent areas within the infiltration.

cause of an injury to the knee, routine chest roentgenograms showed, apart from the sequelae of pleurisy, a walnut-sized, well-defined, rounded, homogeneous mass located peripherally in the anterior upper lobe segment on the left. Laminagrams showed no cavitation or calcifications within this opacity. Laboratory and bronchoscopic findings were normal. On left-sided thoracotomy, a firm, nodular tumor was found superficially situated in the anterior upper lobe segment and it was shelled out with ease. It was grossly as well as microscopically a typical hamartoma. There was an uneventful postoperative course.

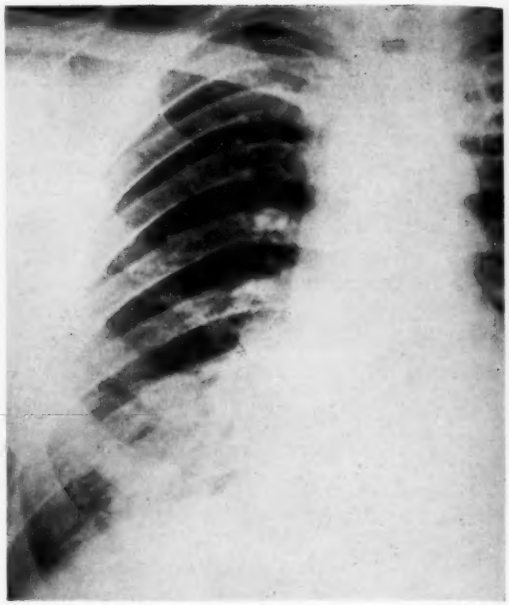

FIG. 4. Case IV. Increased density of the right lower lobe due to an endobronchial hamartoma.

CASE VII (Fig. 7). A male, aged sixty-six, with a history of moderate cardiac symptoms during recent years, but no serious illnesses and no pulmonary symptoms. Routine chest roentgenograms revealed in the left lung field, behind the hilus, basally located in the upper lobe or apically in the lower lobe, a well-defined, somewhat lobular, homogeneous mass the size of a walnut. No cavitation and no calcifications were seen and the pulmonary structures in the surrounding areas were normal. There was some increase in the width of the cardiovascular shadow. Laboratory studies and bronchoscopy showed no abnormalities. Left-sided thoracotomy revealed, basally located in the upper lobe, a round, firm tumor the size of a walnut, which was shelled out easily. On gross and microscopic examination it was found to be a typical hamartoma. There was an uneventful postoperative course.

CASE VIII (Fig. 8, *A* and *B*). A female, aged sixty-one with a history of supravaginal hysterectomy due to menorrhagia at the age of fifty. There had been no other serious illnesses. Owing to mild dyspeptic complaints, as an outpatient roentgen examination of the colon and stomach was done, but it showed no abnormalities. Routine roentgenograms of the lungs revealed, in the right lung field at the site of the anterobasal lower lobe segment, a

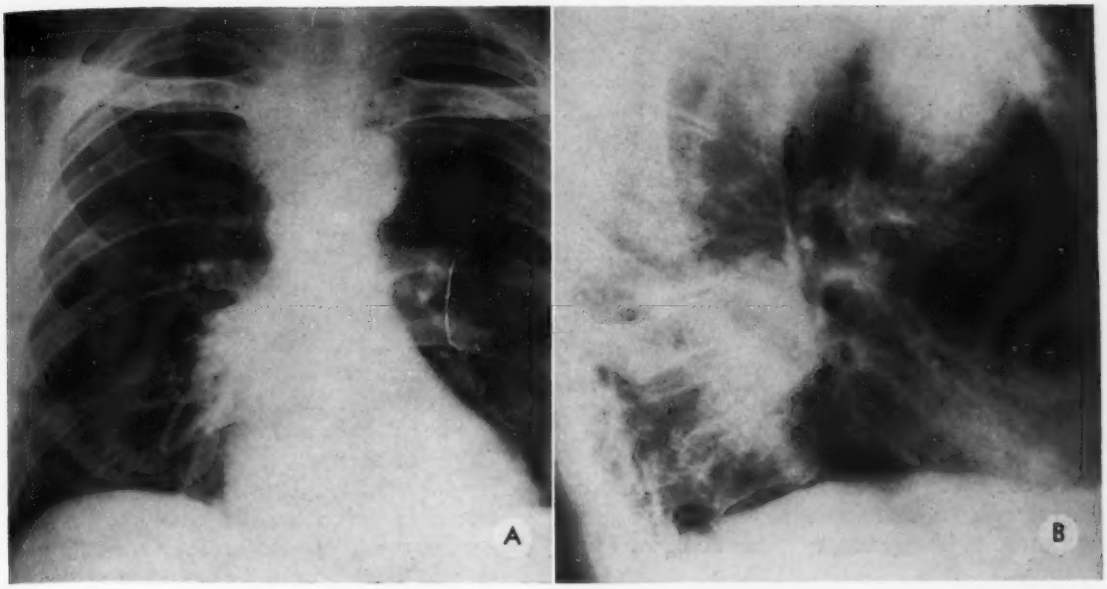

Fig. 5. Case v. (*A*) Frontal view and (*B*) lateral view showing increased density of the apical segment of the right lower lobe due to an endobronchial hamartoma.

well-defined, homogeneous, smooth-margined mass about the size of a hazel nut; surrounded by normal lung tissue. There was no visible cavitation or calcification at the site of the opacity. Laboratory findings were normal. Right-sided thoracotomy revealed in the lower lobe, just below the surface and adjacent to the middle lobe, a well-defined, firm nodule which could be shelled out easily. Gross and microscopic examination showed a typical hamartoma. There was an uneventful postoperative course.

Case ix (Fig. 9, *A* and *B*). A male, aged fifty-seven, who had always been in good

Fig. 6. Case vi. Laminagram showing a well-defined slightly lobulated hamartoma in the anterior segment of the left upper lobe.

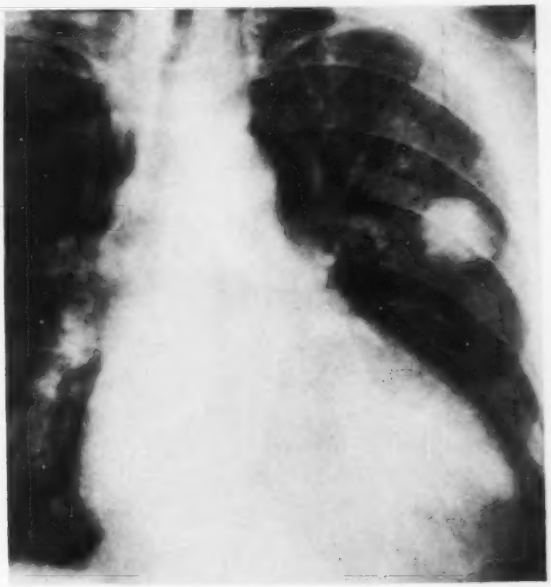

Fig. 7. Case vii. Well-defined, slightly lobulated hamartoma in the left upper lobe.

health. During the two months preceding admission, there had been moderate cough productive of sputum, which had once been blood tinged. There were no other clinical symptoms. Laboratory findings were normal; specifically, no signs of tuberculosis. Bronchoscopy showed no abnormalities. Roentgenograms of the chest demonstrated a homogeneous, well-defined, rounded, somewhat lobulated mass, the size of a hazel nut, located above the right dome of the diaphragm at the site of the middle lobe. The surrounding lung tissue was normal. Laminagraphy did not reveal any cavitation or calcification within the opacity. Right-sided thoracotomy disclosed a firm infiltration, the size of a walnut, close to the pleura. Middle lobectomy was performed and gross as well as microscopic examination revealed a typical hamartoma. The postoperative course was uneventful.

CASE X (Fig. 10, *A* and *B*). A female, aged seven, who had always had a tendency to become short of breath and to catch infectious diseases. During the six months preceding hospitalization she had had pneumonia three times. She was admitted with dyspnea and a new attack of fever. Hemoglobin was 80 per cent; erythrocyte sedimentation rate, 93 mm. Bronchoscopy showed the right main bronchus to be greatly narrowed, not admitting the bronchoscope, but no tumor tissue or foreign body was visible. Roentgen examination of the lungs demonstrated total atelectasis on the right with marked displacement of the mediastinum to the right. Centrally in the right lung around the main bronchus there was a large, irregular patch of calcification. Laminagrams did not show any necrotic cavities. On right-sided thoracotomy, the lower lobe was found to be small and atelectatic. The upper lobe was difficult to distinguish from a nodular tumor which was larger than a closed fist and which adhered to the pericardium inferoanteriorly, to the superior vena cava, and to the pulmonary artery, and was difficult to free. Lobectomy was technically out of the question, so pneumonectomy was performed. Examination of the removed specimen showed that the lower lobe bronchus was free but that it had been completely compressed by the large tumor situated in the upper lobe. The tumor was solid, grayish white, and of uniform structure with diffuse patches of calcification. Microscopic examination revealed that it was a typical hamartoma. The postoperative course was uneventful.

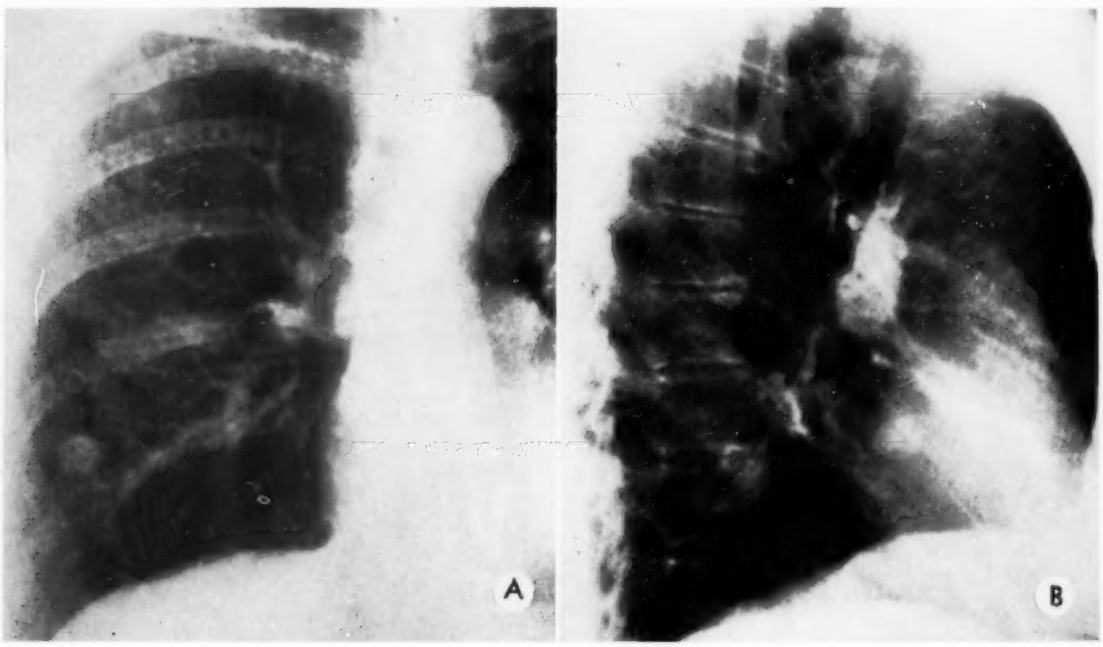

FIG. 8. Case VIII. (*A*) Small, round, well-defined hamartoma without lobulation in the anterior segment of the right lower lobe. (*B*) Lateral view.

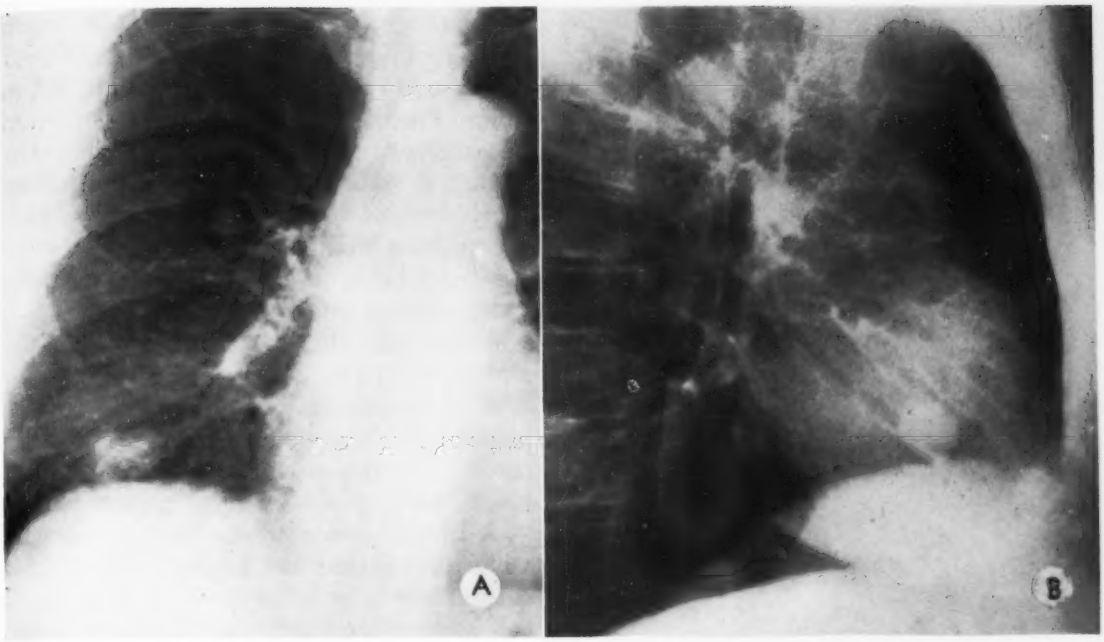

Fig. 9. Case IX. (*A*) Well-defined, lobulated hamartoma in the right middle lobe. (*B*) Lateral view.

COMMENT

Intrapulmonary Hamartomas. Cases II, VI, VII and VIII were of the intrapulmonary type. There had been no clinical symptoms or signs, and all were detected by chance. Case IX also belongs to the intrapulmonary type, but for some unknown reason the tumor had given rise to cough and hemoptysis, which led to its detection. In Case I, which likewise is of the intrapulmonary type, there had been no symptoms and the tumor was detected by chance; however, it is distinguished by its size and by the fact that the patient was only five years of age.

Endobronchial Hamartomas. The remaining four cases were of the endobronchial type. The roentgen findings in Cases III, IV and V were of segmental atelectasis due to obstruction and in Case X of unilateral total atelectasis due to obstruction as well as compression. In the last mentioned case the tumor was invasive, adhering to the pericardium and great vessels, but without any signs of malignancy. All 4 patients had clinical symptoms or signs: cough, expectoration and hemoptysis in 3, dyspnea and recurrent attacks of fever in 1 (Case X).

Only in 1 case (Case X) was calcification found in the hamartoma.

DISCUSSION

Practically all authors up to 1955 report that deposition of calcium is an important diagnostic criterion of pulmonary hamartoma. Lemon and Good[8] were the first to emphasize the uncertainty of roentgenographically demonstrating calcium within the opacity. Bleyer and Marks[2] state frankly that calcification is not of much diagnostic aid, since it is hardly demonstrable in more than about 15 per cent. Of the 15 cases of Bateson and Abbott,[1] calcifications were roentgenographically demonstrated in 2, and in the present series of 10 patients in only 1.

As regards the sex ratio, most authors report a marked male preponderance (Hickey and Simpson[6] 4:1, McDonald *et al.*[9] 3:1, and Carlsen and Kiaer[4] 3.5:1). In Lemon and Good's[8] series of 15 cases, the ratio was 2:1; while in Bleyer and Mark's[2] series and in the present report this pre-

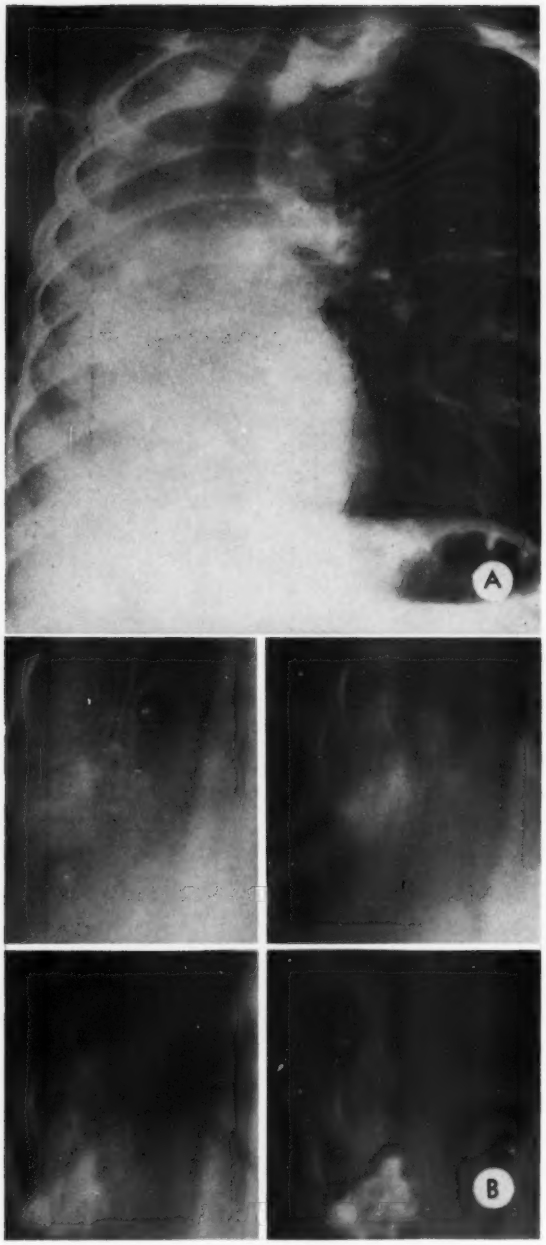

FIG. 10. Case X. (*A*) Total atelectasis of the right lung due to a hamartoma in the upper lobe. The bronchus of the upper lobe is blocked by the tumor and the bronchi of the middle and lower lobes are compressed by it. (*B*) Laminagrams showing pronounced calcifications within the tumor.

ponderance was practically nonexistent (14:11 and 6:4, respectively).

Most authors have mentioned the striking fact that a disease which, judging from all findings, is congenital is uncommon among children and adolescents. Bleyer and Marks[2] even use this fact as an aid in differentiating between tuberculoma and hamartoma, stating that only 4 per cent of hamartomas occur in patients under forty years of age. This is at variance with the present findings, 5 of 10 patients (50 per cent) being under forty and 3 of these (30 per cent) under ten years of age.

As far as the location is concerned, Bragg and Levene[3] reported the distribution to be proportional to the size of the individual lobes, the right lower lobe being most commonly involved. Lemon and Good[8] had a right-sided preponderance of 3:1. In the present series the ratio is 6:4; while Bleyer and Marks[2] had only 6 right-sided cases in 25.

Within the pulmonary tissue proper, the tumor is most often in a fairly superficial, often subpleural location. It is important to bear in mind, however, that the hamartoma may affect any part of the lung, and that a distinction must be made between the type which is remote from and the one which is close to the bronchus, *i.e.*, the intrapulmonary as opposed to the endobronchial type. The latter gives rise to bronchial irritation leading to cough and expectoration and in some cases hemoptysis (Cases III, IV and V). Roentgenologically, the appearance is characterized by atelectasis at the site of the involved area. In the more unusual cases of pronounced obstruction, the clinical signs are dyspnea and recurrent pneumonia (Case X).

The intrapulmonary type is more common, presenting roentgenologically as a roundish, smooth-margined, well-defined, and frequently somewhat lobulated mass (coin lesion) with normal surrounding lung tissue. It seldom gives rise to symptoms, but according to the literature it may at times cause pain, possibly due to pressure upon intercostal nerves (Bateson and Abbott[1]).

In the present series 6 were intrapulmonary. Only 1 of these patients (Case IX)

had clinical symptoms, which were in the form of bronchial irritation causing cough and hemoptysis on one occasion. The explanation is not known.

According to Bateson and Abbott,[1] in a review of the 253 published cases of pulmonary hamartoma, 226 belonged to the intrapulmonary and only 27 to the endobronchial type. This report adds 10 cases, 6 of them intrapulmonary and 4 endobronchial.

SUMMARY

Ten new cases of pulmonary hamartoma are reported. Six were of the more common intrapulmonary type showing the usual roentgen appearance (coin lesion), and only 1 of these 6 patients had clinical symptoms of pulmonary disease. The remaining 4 cases belonged to the very rare endobronchial type, which has been reported previously in only 27 instances. This type is characterized roentgenologically by obstruction atelectasis and gives rise to clinical symptoms and signs, *viz*, cough, expectoration and, in some cases, hemoptysis and recurrent pneumonia. Five of the patients were under forty years of age, 3 of them being under ten years of age.

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CYSTITIS EMPHYSEMATOSA*

19 CASES WITH INTRALUMINAL AND INTERSTITIAL COLLECTIONS OF GAS

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THE presence of gas in the urinary bladder, although stated to be a rare condition, has been the subject of numerous reports in the medical literature.

Cystitis emphysematosa, the presence of gas vesicles in the wall of the bladder, has been arbitrarily distinguished from primary pneumaturia, the presence of gas in the lumen of the bladder, with or without its passage via the urethra. There appears to be no justification for this distinction since they would appear to be different and occasionally concomitant stages of the same basic disease process with common etiologic factors. Despite this, separate and exclusive literatures have developed under each heading, and the essential identity of the two conditions has not been stressed.

PRIMARY PNEUMATURIA

The literature on primary pneumaturia is considerably more voluminous than that on cystitis emphysematosa and dates back to 1671, when, in a publication entitled "Curiosities of Nature" a case was described in which gas was found in the urinary bladder.[28] In 1825 Boismont made reference to spontaneous formation of gas in the bladder,[8] and in 1882 Keyes[12] reported the first 2 cases in the American literature, although these were apparently the result of instrumentation. Senator[25] was the first to propose a classification of causes of gas in the bladder in 1891. He divided cases into three groups: (1) those due to instrumentation, (2) those due to the presence of fistulous connections between the bladder and the bowel, and (3) spontaneous formation of gas in the bladder. In this latter group are the cases with which we are dealing when we speak about "primary" pneumaturia and cystitis emphysematosa, although it is not always possible to distinguish with certainty between group 1 and group 3. In 1898 Kelly and MacCallum[10] reviewed the literature and were able to find 16 cases of gas in the bladder due to fermentation. Nine of the 16 had glycosuria and several of the cases had an associated obstruction of the lower urinary tract. There were no bacteriologic studies of these early cases. Mulsow and Gillies[23] are credited with the first report of a patient in whom the diagnosis of pneumaturia was made roentgenographically.

Between 1898 and 1948 there were many isolated case reports, the majority being in the German literature. In 1948 Arthur and Johnson[1] reported a case in a diabetic female in which *E. coli* was the organism incriminated as the cause of the underlying cystitis. They were able to find 25 cases in the literature with their own being the 26th. Of these only 5 were found in the American literature. Spring and Hymes[27] again reviewed the literature in 1952 and were able to find 38 cases, with glycosuria being present in 21 of these. They tabulated the glycosuric and nonglycosuric cases and showed that the responsible organism in the majority was either *E. coli* or *Aerobacter aerogenes* with inconstant findings of other organisms, including *B. proteus*, *Staph. aureus*, *Streptococci*, *Nocardia*, yeasts and in one case *B. Friedländer*. (It is worthy of note that *Aerobacter aerogenes* and *B. Friedländer* are bacteriologically indistinguishable and are only recognized as different entities by the different pathologic conditions which they produce. It would seem highly likely that the infecting organism in the case cited was, as in many of the re-

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maining reports, *Aerobacter aerogenes* also.) They were likewise able to find a high incidence of urinary retention due to a variety of causes including benign prostatic hypertrophy, cord bladder, and diverticula of the bladder. There was invariably an associated infection. Since that time several other cases have been reported.[2,7,8,17,30] All of these cases were diabetic patients. A further case has been noted by Kent[11] who reported, in addition, 3 cases with gas in the urinary bladder, apparently due to fistulous connections between the bladder and the gastrointestinal tract.

Fineman *et al.*[7] were able to find only 4 references to the roentgenologic diagnosis of primary pneumaturia. Boijsen and Lewis-Jonsson[2] were the first authors to hint at the identity of primary pneumaturia and cystitis emphysematosa. Their paper contains references to the literature under both headings and the roentgenograms reproduced in their paper clearly show, as do roentgenograms reproduced in other papers on cystitis emphysematosa, the presence of gas both in the lumen of the bladder and within the bladder wall. Thomas and Sandler,[30] in reporting their case, state that cystitis emphysematosa occurs only rarely with pneumaturia.

CYSTITIS EMPHYSEMATOSA

The condition "cystitis emphysematosa" was originally described as a pathologic entity only. Hueper,[9] in 1926, described the changes of cystitis emphysematosa in a female dog which was submitted to him for autopsy. The dog proved to have glycosuria, hyperglycemia and changes in the pancreas which included complete absence of islets of Langerhans. No bacteriologic studies were made. He described an additional 11 clinical cases collected from the literature, but there are few clinical data on these cases and there were apparently no adequate urinary studies. To Mills[18–21] goes the credit for giving excellent pathologic descriptions in a series of 12 cases reported in 1930. These were all seen at autopsy. Originally, he was uncertain as to whether the findings represented postmortem changes or results of disease in the living subject. However, he eventually came to the conclusion that the disease was a clinical entity and that it would ultimately be demonstrated in a living subject. None of Mills' patients were diabetics, but he pointed out that 2 of his subjects had had repeated intravenous glucose infusions. The presence of glycosuria in these 2 patients was not stated. His predictions regarding the discovery of cystitis emphysematosa in a living subject were shortly confirmed by Ravich and Katzen,[24] who reported a case in a living patient with recovery. Following this, several observers noted the condition on cystoscopic examination of the bladder. Burrell[4] gave a description of the cystoscopic appearances in a nondiabetic and was able to reproduce the condition by means of an ingenious experiment. He produced a chemical cystitis by means of silver nitrate in a male rabbit made glycosuric by the intravenous infusion of glucose and then inoculated the bladder with the responsible organism (*E. coli*) from a patient having cystitis emphysematosa. The rabbit developed the typical changes in the bladder wall. Other descriptions of cystoscopic findings were given by Levin,[14] Lund *et al.*,[15] Marquardt,[16] and Burns.[3] Lund *et al.* were able to collect gas from the vesicles and found it to be carbon dioxide.

Wells,[31,32] in 1938, reported 7 cases of cystitis emphysematosa. Only one case had glycosuria and he stated that he could find that neither glycosuria nor trauma were significant etiologic factors. He also described the pathologic findings in a subsequent paper. In 1949, Teasley[29] reviewed the 43 cases in the literature and added one case of his own, a diabetic patient. Seven of these were in diabetics. He cited infection with *E. coli* and hyperglycemia as common findings (although sufficient bacteriologic and urinary data were not available on many of the earlier cases, as previously noted). Since that time cases have been reported by Milner *et al.*,[22] Faingold *et al.*,[6] Zatzkin,[33] Lane and Francke,[13] and Sotero-

poulos *et al.*[26] Faingold and co-workers'[6] 4 cases all showed glycosuria, 2 of them being confirmed diabetics, the remaining 2 having glycosuria at some time during the course of their illness but with diabetes not definitely established. Three of these had *E. coli* as the infecting organism and the diagnosis was made roentgenologically in all 4 cases. They pointed out that the condition was transitory in nature and offered this as an explanation for its apparent rarity as a diagnosis. Zatzkin's[33] case was in a patient who had glycosuria associated with hemochromatosis. The infecting organism was not stated. The case of Soteropoulos *et al.*[26] was in a diabetic and is of interest as being the first reported case in which the vesicles were seen to extend into the distal third of both ureters. They cultured *E. coli* and *Aerobacter aerogenes* from the urine.

In all, there appear to be 52 cases in the literature under the heading of "cystitis emphysematosa." Of these, 13 were known to have glycosuria. Data in some of the remainder are incomplete. *E. coli* is the common denominator from a bacteriologic standpoint. Under the heading "primary pneumaturia," there appear to be 46 other cases in the literature, 29 of which had glycosuria. We are reporting an additional group of 19 cases (Table I and II).

The increasing frequency of diagnosis is apparently due to the increasing awareness of clinicians and radiologists of this condition.

It is the impression of the author that the literature on each subject should now be integrated. Findings common to the two conditions as seen in the many reported cases include (1) high incidence of glycosuria in the presence of urinary stasis and obstruction, (2) infection with *E. coli* or *Aerobacter aerogenes*, and (3) production of gas in the wall of the bladder, in the bladder lumen, or in both places.

CASE MATERIAL

During the twelve year period from 1949 through 1960, 19 patients have been studied in the radiology department of the New England Deaconess Hospital with evidence of gas either in the wall or the lumen of the urinary bladder or in both places. Fifteen of these cases have been seen in the last three years.

The 19 cases comprise 15 diabetic and 4 nondiabetic patients. Five of the 6 male patients and 10 of the 13 female patients suffered from diabetes. In 3 cases, 1 male and 2 female, episodes of cystitis emphysematosa occurred on two separate occasions with evidence of persisting urinary tract infections in the intervening period.

ILLUSTRATIVE CASE HISTORIES

CASE 3. B.S. This twenty-five year old white female was admitted to the New England Deaconess Hospital for the twelfth time on October 22, 1958, her previous admissions being connected with the complications and control of her diabetes which had been diagnosed fifteen years previously. She had been fairly well until the day of admission when she experienced gross hematuria and crampy pain in the left flank radiating into the left groin. She vomited repeatedly.

Physical examination: Well developed and well nourished. Pulse 76, temperature 99° F.; heart and lungs were normal. The popliteal pulses were not palpable. There was marked tenderness in the left costovertebral angle and also in the left lower quadrant.

Urinalysis: Grossly bloody specimen, albumin 65 mg. per cent, glucose 2.5 gm. per cent and there were 80 to 100 white blood cells per high power field. The nonprotein nitrogen was 55 mg. per cent. Urine culture showed *Aerobacter aerogenes*.

An admission roentgenogram of the abdomen showed gas in the lumen and in the wall of the bladder. An intravenous pyelogram revealed evidence of right sided chronic pyelonephritis and a questionable cyst on the left kidney together with an estimated 6 ounces of residual urine.

Course: She was treated with achromycin, sedatives and analgesics and her diabetes was brought under control. On this regimen her symptoms subsided and she was discharged on November 4, 1958. She was again admitted with a urinary tract infection and gross hematuria on November 4, 1959. On this admission a retrograde pyelogram revealed filling defects in

Table I

PERTINENT FINDINGS IN CASES STUDIED

Case No.	Age and Sex	Diabetes (yr.)	Associated Conditions	Organism	Urine Data Reaction	Urine Data Glucose	Red Blood Cells	White Blood Cells	Remarks
1 A.B.	63F	10	Diabetic neuropathy; horseshoe kidney; arteriosclerotic hypertensive heart disease	*E. coli*	acid	o-tr.	o	60–80	Minimal glycosuria
2. M.B.	72F	—	Cystocele; bedridden 20 yr.; Menière's disease 20 yr.; frequency and spasmodic incontinence many years	*Staph. aureus*	acid	o	10–20	10–20	On catheter drainage of bladder; low bladder capacity of 180–200 cc.
3 B.S.	25F	15	Retinopathy; nephropathy; neuropathy; degenerative vascular disease; chronic pyelonephritis	*Aerobacter aerogenes; B. proteus*	1. acid 2. acid	+ +	gross o	80–100	Two separate episodes of cystitis emphysematosa
4 M.M.	44F	—	Bilateral narrowing of renal arteries on aortogram; small right kidney	—	alkaline	o	o	o	Catheterized two weeks prior to intravenous pyelography; no cystoscopy; had splenorenal shunt operation for narrowing of left renal artery
5 E.F.	36F	14	Diabetic nephropathy and retinopathy	*E. coli*	acid	o	o	25–45	"Tests very good" for few days prior to admission
6 C.N.	26F	14	Diabetic neuropathy; nephropathy and retinopathy with residual urine of 400 cc.	*E coli*, *Aerobactor aerogenes*	alkaline	+	gross	—	Volunteered that she had passed gas in urine; had had poor diabetic control with frequent high blood sugar readings
7 A.O.	77F	35	Pernicious anemia 12 yr.; aortic stenosis	*E. coli*	acid	+	gross	100–125	Gas observed during fluoroscopy for gastrointestinal series; died during a subsequent admission with *E. coli* septicemia
8 M.D.	64F	22	Chronic urinary infection; diabetic retinopathy	*E. coli*	acid	+	gross	??	Passed gas in urine on 2 or 3 occasions; much residual urine; *E. coli* isolated was a marked gas producer; readmitted 2 yr. later with gas in bladder
9 A.R.	55F	—	Urinary tract infection 37 yr.	*E. coli*	alkaline	o	o	—	Retrograde pyelography 7 da. previously; bladder obscured by rectal contents at that time
10 A.K.	80M	13	Diabetic neuropathy; cardiac decompensation	*E. coli*	acid	o	o	40–60	Little or no glycosuria on admission in spite of a blood sugar of 275; died from cerebrovascular accident; no autopsy or urine data on last admission
11 A.S.	61F	2	Cardiac decompensation led to admission; hematuria developed after admission; diabetic neuropathy	*E. coli, Strep. anhemolyticus*	?	?	gross	?	Fairly large residual urine of 250 cc.; cystitis emphysematosa cleared in 4 days
12 J.B.	61M	8 mo.	Large prostate; prostatectomy; postoperative pneumonia	*Aerobacter aerogenes*	acid	+	gross	10–20	Trabeculation and stasis in bladder probably on basis of prostatic enlargement; prostatectomy with postoperative pneumonia; agranulocytosis; autopsy
13 M.M.	67F	13	Hypertension; diabetic neuropathy	*E. coli*	acid	+	o	80–100	
14 C.T.	83M	—	Gastric ulcer treated surgically; colonic and bladder diverticula	*E. coli; Aerobacter aerogenes; Staph. aureus*	alkaline	o	occasional	250–75	Gastrointestinal studies showed gas probably in diverticulum prior to cystoscopy; definite gas in diverticulum after cystoscopy; no evidence of fistulae on cystoscopic examination
15 E.L.	54F	23	Arteriosclerotic heart disease; neuropathy; hiatus hernia; "nephrotic syndrome"; 24 previous admissions	*Aerobacter aerogenes*	alkaline	o	occasional	150–175	Persistent *E. coli* urinary tract infections on subsequent admissions
16 C.B.	61M	6	Neuropathy; previous transurethral resection	*E. coli; B. proteus*	?	o-tr.	gross	??	Occasionally passed gas in urine over 2 yr. period but this fact had to be elicited by direct questioning
17 B.D.	23M	10	Diabetic retinopathy; neuropathy; nephropathy	*Aerobacter aerogenes*	acid	+	occasional	100–125	Gas demonstrated in bladder at outside hospital 1 yr. previously
18 F.H.	53M	26	Old tuberculosis treated by surgery; neuropathy; ?retinopathy	*Aerobacter aerogenes*	acid	+	occasional	30–45	Persistence of gas for 1 wk. associated with persistent *Aerobacter aerogenes* in urine; gas disappeared after 14 da.; urine sterile
19 K.B.	27F	12	Bilateral cataracts; bladder neuropathy; severe degenerative vascular disease	*E. coli*	alkaline	+	occasional	30–40	Calcified pelvic vessels; seen 2 yr. later; no gas present in urinary tract

TABLE II

ROENTGEN FINDINGS IN 19 CASES WITH GAS IN THE BLADDER

Case No.	Diagnosis of Gas		Time to Clear	Manifestation	
	Clinical	Roentgenologic		Intramural	Intraluminal
1	o	+	Less than 5 da.	+	+
2	o	+		−	+
3	o	+		+	+
	o	+		±	+
4	o	+		−	+
5	o	+		+	−
6	+	+		+	+
7	o	+		±	+
8	+	+	4 da.	−	+
9	o	+		+	+
10	+	+	4 da.	+	+
11	o	+		+	+
12	o	+	Less than 2 wk.	+	−
13	o	+		−	+
14	o	+		−	+
15	o	+		−	+
16	o	+		±	+
17	+	+	4 da.	+	+
18	o	+		+	+
19	o	+	4 to 5 da.	+	+

the renal pelves (? blood clot) but no evidence of gas in the bladder. She was treated with bed rest, antibiotics and control of her diabetes. The symptoms again subsided. A culture of the urine at this time again revealed *Aerobacter aerogenes*.

On her fourteenth admission on January 25, 1960, she complained of chills and fever, which had been attributed to an upper respiratory tract infection. Physical examination was essentially negative except for blood pressure of 160/110 and pulse of 104. Diabetic retinopathy was noted. On January 27, a roentgenogram of the abdomen showed air-fluid levels in the bladder together with a slight degree of intestinal ileus. An intravenous pyelogram four days later showed evidence of bilateral pyelonephritis in the form of slightly dilated calyces. Bubbles of gas were seen in the bladder and there was residual urine of approximately 6 ounces. Urine culture revealed *B. proteus*. She was treated with insulin, bed rest and chloromycetin, whereupon her symptoms subsided and she was discharged improved on February 3.

Comment. A typical case of primary cystitis emphysematosa with evidence of gas both in the lumen and in the wall of the bladder in the presence of diabetes and stasis, due, in this case, to diabetic neuropathy. Infection of the urine with *Aerobacter aerogenes* was demonstrated on several occasions. It seems possible that the *B. proteus* organism cultured on the last admission may have masked the presence of an infection with another gas-forming organism. It is of interest that the condition recurred after treament and on each occasion responded without undue local sequelae.

CASE 7. A.O. This seventy-seven year old white female diabetic of thirty-five years' duration was admitted to the New England Deaconess Hospital for the twelfth time on June 20, 1958. She had been under treatment for the past twelve years for pernicious anemia in addition to her diabetes mellitus.

She complained of dysuria which had been of fairly recent onset and she had noted gross blood in her urine which was also cloudy. Previous admissions had been in connection with her diabetic control and treatment of her anemia. On one occasion there had been noted a high blood sugar in the absence of glycosuria and on a further occasion bacteruria had been observed.

Physical examination: Not remarkable except for a Grade IV systolic murmur at the apex. Blood pressure was normal and the lungs were clear.

Urinalysis: Specific gravity of 1.005, acid reaction, albumin 19 mg. per cent, glucose 0.3 gm. per cent and 80 to 120 white blood cells per high power field. Cultures of the urine revealed *E. coli*. On the eighth hospital day, gas was observed in the bladder during examination of the gastrointestinal tract. This was confirmed on an intravenous pyelogram two days later which showed good function of each kidney, an air-fluid level in the bladder and a residual urine of greater than 6 ounces.

Course: The patient was treated with chloromycetin and insulin on which regimen she showed marked improvement and was discharged on July 1, 1958. She was re-admitted on December 11, 1958 complaining of "weakness" for two days prior to admission. Thirty-six hours prior to admission she had vomited bile-stained material and twelve hours after this had experienced the onset of generalized

upper abdominal pain with diarrhea. This had continued up until the time of her admission.

Physical examination on this occasion revealed a blood pressure of 60/40 with pulse of 100 per minute and temperature of 99 degrees. The remainder of the examination was unremarkable except for the previously noted cardiac murmur and slight left upper quadrant tenderness.

Laboratory findings: Hemoglobin 11.9 gm. per cent, white blood cells 24,700 rising later to 70,700 with 34 per cent band forms. Serum electrolytes were normal. Blood sugar was 294 mg. per cent. A blood culture disclosed *E. coli* as did a culture of the urine. Urinalysis revealed 20 mg. per cent of albumin, 40 red blood cells per high power field and 10 to 12 white blood cells per high power field. She was treated with intravenous fluids and sympathomimetic amines, but she developed pulmonary edema and died approximately twelve hours after admission.

Autopsy findings: Cultures of the heart blood, spleen and kidney grew *E. coli*. The kidneys showed scattered yellow nodules in both the cortex and the medulla, the nature of which was not clear. There was no evidence of diabetic nephropathy. The bladder wall was thickened, measuring up to 0.8 cm., with a somewhat firm, polypoid, mottled red surface. Microscopically, the bladder showed submucosal infiltration with chronic inflammatory cells, dilatation of blood vessels and occasional hemorrhages.

Comment. This patient is of interest because she had shown on one occasion discrepancy between the blood sugar and the urine sugar, a feature of cystitis emphysematosa which has been mentioned by previous authors.[27] Stasis was also present, presumably on the basis of diabetic neuropathy. She subsequently succumbed to a septicemia with the same organism which was responsible for her bladder infection.

Case 8. M.D. This sixty-four year old white female was admitted to the New England Deaconess Hospital for the first time in January of 1958 with a urinary tract infection (*Aerobacter aerogenes*) which was successfully treated with penicillin and streptomycin. She was discharged, but was re-admitted in July, 1958 with gangrene of the toes of the right foot for which a transmetatarsal amputation was performed. At the time of her admission on November 19, 1958 she had had diabetes mellitus for a total of twenty-two years. She complained at this time of intermittent painless hematuria of six days' duration with passage of gas in the urine on two or three occasions. There was frequency of urination, every two to three hours by day and two to three times during the night.

Physical examination: Well nourished woman, not in great distress. The temperature was 99° F., pulse 90, and blood pressure was 135/60. Heart and lungs were negative. Abdominal examination gave the sensation of a suprapubic mass. Ankle jerks were absent. Diabetic retinopathy was noted.

Urinalysis: Specific gravity 1.010, acid reaction, albumin 470 mg. per cent, glucose 2.2 gm. per cent, and the specimen was "loaded" with red blood cells. Culture of the urine revealed a strain of *E. coli* which produced considerably more gas than a strain of *E. coli* selected at random from those under culture in the laboratory, both growing in a glucose medium. On the second hospital day, an intravenous pyelogram disclosed gas in the bladder lumen which persisted after voiding.

Course: The patient was treated with achromycin and insulin under which regimen her symptoms subsided. However, she was readmitted one year later at which time she again had an *E. coli* urinary tract infection, but with no evidence of gas in the bladder. A further re-admission in November, 1960 showed the bladder to be distended with gas almost to the level of the umbilicus and an infection with *E. coli*. This was again treated with antibiotics. A roentgenogram of the abdomen one month later showed it to be distended but with no gas present.

Comment. This is the only case in the series in which the propensity of the infecting organism for gas production was tested. It might well be that others, if tested, would have shown this property.

Case 12. J.B. This sixty-one year old white male was admitted to the New England Deaconess Hospital for the first time on December 13, 1957 with a six to eight month history of polydypsia and polyuria. Glycosuria and acetonuria had been discovered four days prior to admission. Eight hours prior to admission he began to void blood-stained urine. The past history was essentially negative. Physical examination: Male in acute distress with marked urgency of urination. Heart and lungs were

normal. There was diffuse suprapubic tenderness. The prostate gland was enlarged. The remainder of the examination was essentially negative.

Urinalysis: Specific gravity 1.015, acid reaction, albumin 152 mg. per cent, glucose 1.3 gm. per cent, 70 to 90 red blood cells per high power field, 19 to 20 white blood cells per high power field and bacteria 4 plus. Culture of the urine on two occasions revealed *Aerobacter aerogenes*. The hemoglobin was 13.4 gm. per cent, white blood cells 12,900 with a shift to the left. Blood sugar was 94 on admission and 214 on the second hospital day. On the third hospital day, an intravenous pyelogram revealed trabeculation of the bladder dome with questionable gas vesicles within the wall of the bladder. Re-examination of the pelvis after careful preparation of the rectum on the following day showed positive evidence of gas bubbles in the wall of the bladder.

Course: The diabetes was controlled on 35 units NPH insulin. On the sixth hospital day a cystoscopy revealed a bladder of limited capacity with multiple ecchymotic areas on the posterior wall. At the junction of the floor and posterior wall, there were hyperemic rugations with tiny, gray cysts. A biopsy of the bladder wall was reported as showing chronic inflammation and fibrosis. Subsequently, he developed a postoperative pneumonia and both this and his urinary tract infection proved refractory to all antibiotics which included chloromycetin, gantrisin, achromycin, polymyxin, furadantin, streptomycin, triple sulfa and albamycin. Following this he developed agranulocytosis and succumbed to a septicemia on February 9, 1958. An autopsy showed an organizing pneumonia of the right middle lobe with terminal aspiration and pulmonary edema. A swab from the prostate gland showed *E. coli* and a swab from the bladder revealed *Pseudomonas* and *Streptococcus hemolyticus*. Examination of the bladder showed it to be markedly hemorrhagic with papillary projections around the area of entrance of the indwelling catheter. In addition, this area was markedly friable. On the posterior wall just above the trigone, there was an almost perfectly round area of denudation of the mucosa with surrounding radial puckering of the wall. The whole of the bladder had a "boggy" consistency. The prostate gland showed many thick walled abscesses up to 1.5 cm. in diameter. Multiple venous thromboses were present. Microscopic examination of the bladder showed acute and chronic inflammatory cells. There were nests of normal mucosal cells below the level of the rest of the mucosa plus a fibrous, healing ulcer. A necrotic area containing clumps of bacteria was situated near the bladder dome and was surrounded by congested vessels, occasional hemorrhages, and a zone of reaction composed mostly of eosinophils, lymphocytes and monocytes.

Comment. This is a case of cystitis emphysematosa with cystoscopic findings. Persistent infections in the bladder region and a complicating postoperative pneumonia with the exhibition of multiple antibiotics in an attempt to control these infections apparently resulted in agranulocytosis and death from septicemia. Autopsy also showed severe prostatic infection, but it is not clear whether the prostatic infection preceded the bladder infection or vice versa. The cystoscopic appearance corresponds fairly closely with that previously noted by Mills[18-21] and others.

FINDINGS

The pertinent clinical, roentgenologic and laboratory data of the 19 cases studied are summarized in Tables I and II. There was a striking preponderance of females to males in an approximate ratio of two to one.

The age of the patients ranged from twenty-three to eighty-three years. Of the 15 diabetic patients, the average duration of diabetes was 14.2 years with a range of a "few months" to 35 years. The salient clinical findings were a high incidence of glycosuria and evidence of urinary tract infection, which was often recurrent. Gross hematuria was a common finding in cases in which gas in the bladder wall was a feature. Subsequent histologic examination in 2 such cases showed hemorrhage within the bladder wall. The responsible organism in almost every instance was either *E. coli* or *Aerobacter aerogenes*, the two exceptions being Case 3 and 4. In the former case the organism cultured during one of her two episodes of cystitis emphysematosa was

B. proteus, an organism noted for tending to "swarm" over other organisms growing with it. In the other case the culture of the urine showed "no growth." This patient had, however, been on antibiotic therapy shortly before the diagnosis was established.

Diagnosis was made by roentgen examination alone in 15 cases and by both roentgenograms and clinical history in the remaining 4 cases. Gas in the wall of the bladder was the sole manifestation in 2 cases; gas in the lumen of the bladder was the most prominent feature in a further 6 cases, and the remaining 11 cases had evidence of gas in both the wall and the lumen of the bladder. Only 2 of the patients who had gas present in the lumen of the bladder volunteered the information that they had passed gas when voiding. One further patient gave this information on direct questioning after gas was discovered by roentgen examination.

It is to be noted that 4 of the diabetic patients had negative tests for urine glucose on admission.

Associated findings included a high proportion of chronic urinary tract infections and urinary stasis. Stasis in the diabetic patients was evidently due chiefly to bladder neuropathy. In the 6 cases of which follow-up roentgenograms were available, the time taken for the gas to disappear under treatment was less than four days in 4 cases, and an indeterminate period less than two weeks in another case. Persistence of gas in the remaining case was associated with persistence of the infecting organism in the urine despite the antibiotic treatment and control of diabetes. The urine was acid in reaction in 10 cases, alkaline in 4 (including 3 of the 4 nondiabetic patients) and in a further case this information is not present in the laboratory data.

Of the 2 cases which came to autopsy, the findings in the bladder were those of an acute and chronic submucosal inflammatory reaction. One of the cases was also cystoscoped prior to death and the bladder showed an intense hemorrhagic reaction with vesicles and ulcerated areas in the mucosa.

ROENTGEN FEATURES

The gas within the bladder wall or lumen, which is the essential feature of this condition, must be distinguished from gas in the rectum and sigmoid. This is usually not difficult, but, if necessary, roentgenograms of the bladder region with the patient in the erect position must be taken to demonstrate the characteristic air-fluid level (Fig. 3, *A* and *B*). The gas is usually visible on plain roentgenograms taken in the supine or erect position. An intravenous pyelogram is not essential in order to make the diagnosis but gives valuable additional information regarding contributory factors (Fig. 4, *A* and *B*). It may show prostatic hyperplasia with obstruction or a large residue resulting from diabetic neuropathy.

When the gas is situated chiefly in the wall of the bladder, diagnosis presents no difficulty since the appearance is quite characteristic. It consists of a narrow radiolucent line of gas bubbles outlining the bladder wall when seen in profile and a collection of vesicles varying in size from a few millimeters up to one centimeter and having a "cobble-stone" appearance when seen "*en face*." In most of these latter cases a collection of intraluminal gas is also present (Fig. 1, *A* and *B*). Persistence of gas after a few days of treatment is invariably associated with persistence of the infection.

In those cases where intraluminal gas is the predominant feature, careful inspection will usually reveal the presence of a few bubbles within the tissues of the bladder wall (Fig. 2, *A* and *B*).

DISCUSSION

It would appear that cystitis emphysematosa is a condition which is not so rare as many previous reports would indicate. Experience in this department of radiology would seem to suggest that it is increasing in frequency since we have seen 15 cases in the last three years as compared to only 4

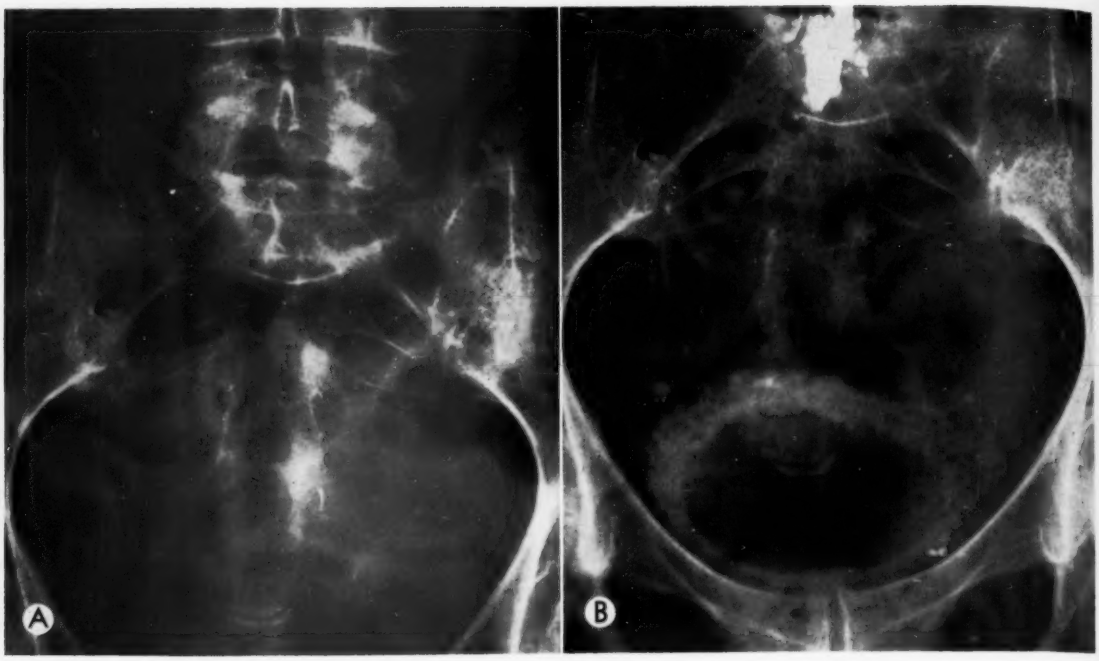

FIG. 1. Case 1. (*A*) Plain roentgenogram of the bladder region before discovery of cystitis emphysematosa. The bladder is moderately distended but shows no evidence of gas. (*B*) Two weeks later. Characteristic appearance with vesicles seen in profile and "*en face*." Note also the collection of gas within the lumen of the bladder.

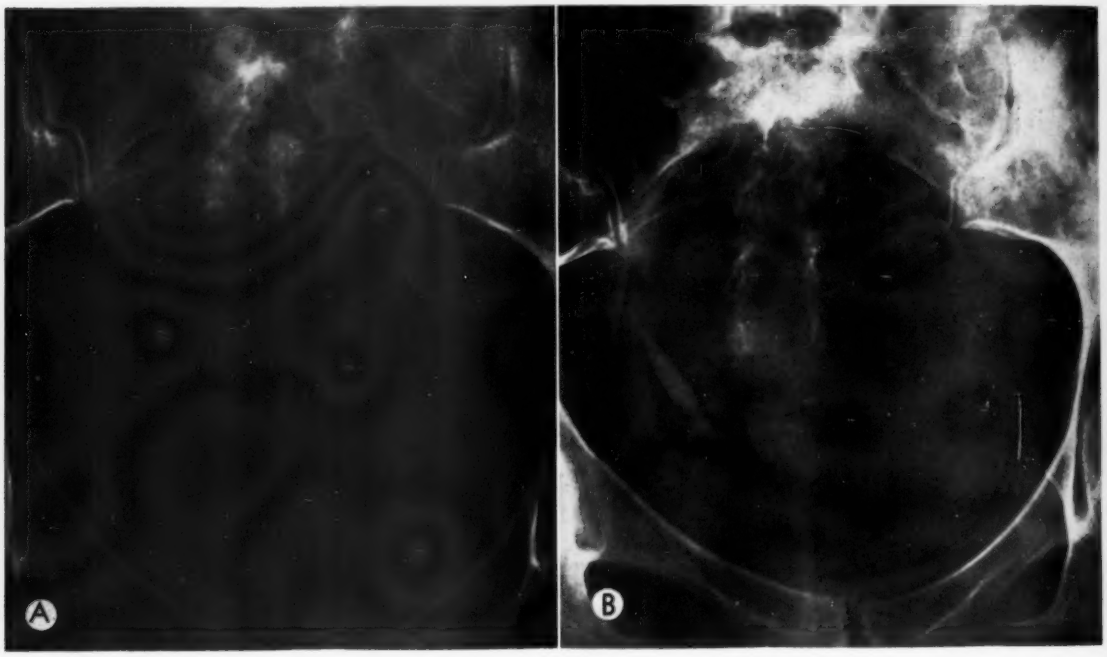

FIG. 2. Case 6. (*A*) Roentgenogram showing gas mainly within the lumen of the bladder. (*B*) Several minutes later after voiding most of the gas. The bladder wall can now be distinguished by the interstitial vesicles which were previously obscured by the larger collection of intraluminal gas.

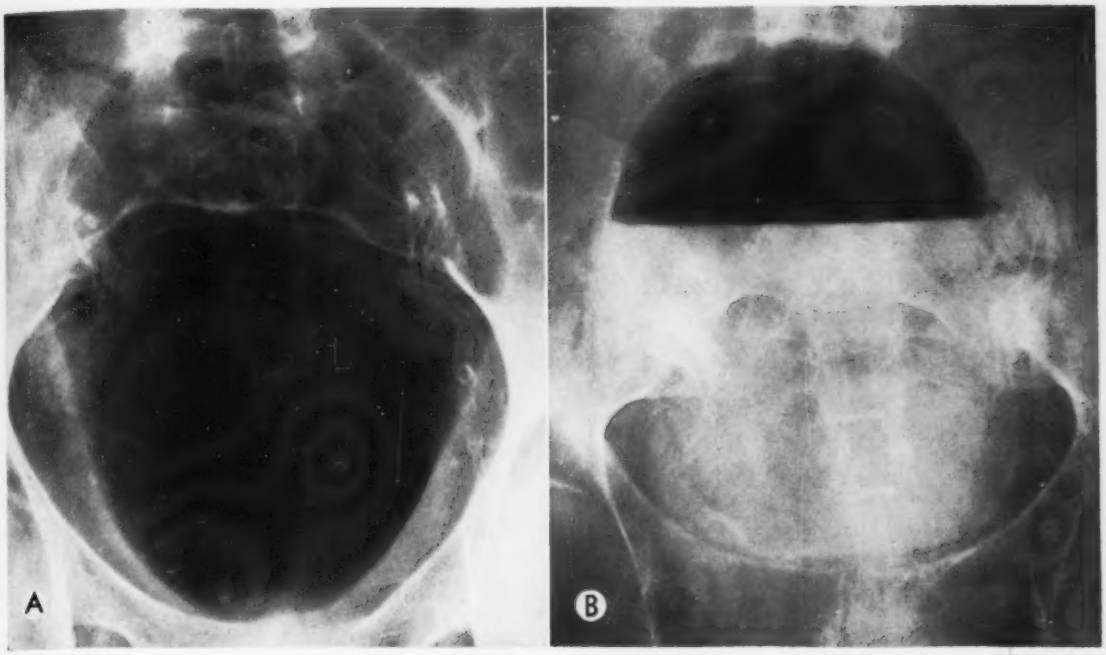

Fig. 3. Case 8. (*A*) Roentgenogram showing the distended gas-filled bladder extending almost to the level of the umbilicus. (*B*) Erect roentgenogram showing the characteristic air-fluid level.

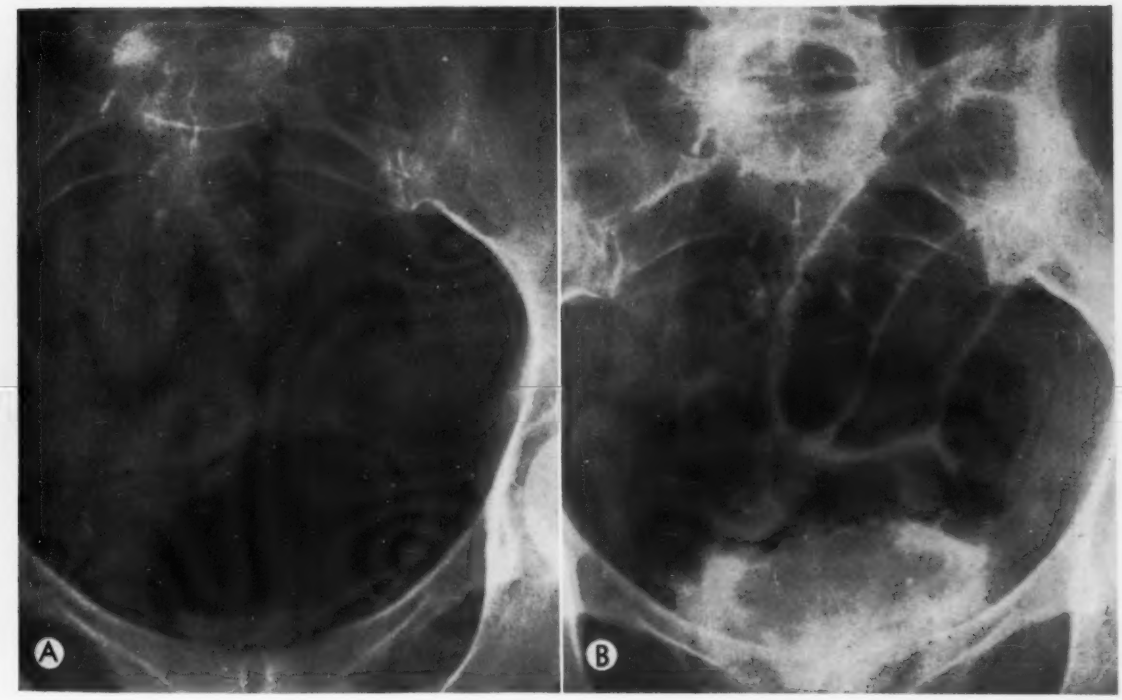

Fig. 4. Case 11. (*A*) Plain roentgenogram prior to intravenous pyelography. Characteristic interstitial vesicles and intraluminal gas bubbles are seen. (*B*) Intravenous post-voiding pyelogram showing slight depression of the bladder base and residual urine.

cases in the previous eight years. It should be noted that our hospital population of approximately 10,000 patients per year includes at least 3,000 diabetics. The proportion of diabetic admissions has remained relatively constant, although the incidence of patients with cystitis emphysematosa has apparently increased. It is noteworthy, in this respect, that in the series of necropsy cases reported by Mills,[20] in 1930, glycosuria was not a conspicuous feature. Nevertheless, in the recent literature most of the reported cases have had glycosuria, usually due to diabetes mellitus, as a notable feature of the disease, and, in general, the incidence of glycosuric cases in the early literature, although much higher than in the general population, was not so high as in cases reported during the last ten years. It is possible to explain this on several grounds. First, glycosuria is not an essential prerequisite for the development of gas in the bladder. However, infection is definitely necessary. Chronic, refractory urinary tract infections were much more prevalent in the pre-antibiotic era and all of the nondiabetic cases reported here have had evidence of long standing urinary infections. Since the advent of antibiotic therapy, moreover, patients with diabetes mellitus have formed a hard core of cases in which urinary tract infections are, because of the glycosuria, difficult to control, and when apparently controlled tend to recur readily. Similar infections in nondiabetic patients would be more amenable to complete eradication. Secondly, at the time of the early reported cases, the detection and adequate control of diabetes was less advanced than at present and the longevity of diabetic patients was not nearly so great as now. Patients therefore did not live to develop the late complications, such as diabetic neuropathy involving the bladder which has been such a conspicuous and recurring feature of the cases presented here. Furthermore, the roentgen examination has led to the detection of many cases of cystitis emphysematosa, both in glycosuric and nonglycosuric patients which would otherwise have been overlooked. The fact that this condition is relatively transient has been stressed by Faingold *et al.*,[6] and is confirmed in this series; in 15 of our cases the diagnosis was unsuspected clinically and in the 6 cases with follow-up roentgenograms at least 4 showed disappearance of the gas within four days.

One of the present cases lends support to the thesis that, in addition to glycosuria, it is necessary to have an organism which has increased propensity for gas formation. The organism isolated from Case 8 showed marked increase in gas production in glucose over a randomly selected strain of a similar organism.

That the condition is a relatively benign one is evidenced by the fact that none of the patients in the present series died as a direct result of the local condition. One patient died indirectly as a result of the treatment of this and other infections, and another patient died remotely as a result of an *E. coli* septicemia in which the chronic bladder condition could probably be incriminated etiologically. Cystitis emphysematosa will, on occasion, clear merely by treatment of the glycosuria and without the administration of antibiotics. In glycosuric cases the mechanism of gas formation is believed to be due to fermentation of glucose with formation of carbon dioxide, together with butyric and lactic acid. All of our diabetic cases except 2 showed an acid urine, whereas 3 of the 4 nondiabetic cases showed an alkaline urine. In the nondiabetic cases, the production of gas is thought to be due either to the breakdown of an abnormal albumin in the urine or else to the presence of a strain of *E. coli* which is capable of acting on normal albumin.

Clinically, the disease is of importance since the fermentation of glucose leads to a false low reading on urine testing, which therefore does not accurately reflect the blood sugar levels. This can lead to a vicious cycle of poor control, higher urine glucose levels, further infection leading onto further fermentation. Since the patient is infrequently aware of pneumaturia or

rarely reports it, the radiologist may be the first to break the cycle.

Finally, it is important to stress the fact that the presence of gas in the wall of the bladder and gas in the lumen of the bladder are manifestations of the same underlying etiologic factors. It has not been possible to determine from the cases studied here whether the gas in the lumen of the bladder is derived from rupture of vesicles or whether it is generated in the lumen itself. It may well be that both mechanisms are in operation at the same time. It would seem that, in order to avoid confusion, the condition should be referred to as "cystitis emphysematosa" and that the term pneumaturia, which is merely a description of a symptom, should be abandoned.

SUMMARY

The literature on "cystitis emphysematosa" and "pneumaturia" has been briefly reviewed. Findings in 19 additional cases are presented.

The high incidence of glycosuria and infection with gas-producing coliform organisms (*E. coli* and *Aerobacter aerogenes*) is stressed, together with the presence of urinary stasis.

The diagnosis is readily overlooked clinically and is essentially a roentgenologic one. The importance of the condition with regard to the control of diabetes is emphasized.

"Cystitis emphysematosa" and "pneumaturia" would appear to be manifestations of a single problem and it is proposed that the single term "cystitis emphysematosa" should be used to avoid confusion.

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BULLOUS EDEMA OF THE BLADDER SIMULATING TUMOR*

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MANY pathologic conditions of the urinary bladder may be suggested by the appearance of the bladder in excretory urography. These include diverticula, extravasation, stones, trabeculation, abnormal capacity, and tumors. There are also many filling defects simulating tumor, such as those caused by foreign bodies, soft radiolucent stones, blood clots and "catheter edema." The latter, edema of the bladder dome caused by continued contact with a Foley catheter tip and bag, is a relatively common finding. However, a recent review of the American literature and of several well known urologic texts discloses no mention of this entity.[1–6]

Patients on constant Foley catheter drainage maintain an empty bladder, thus causing the catheter tip and bag to contact the posterolateral and superior surface of the bladder. This may lead to localized bullous edema which, when seen on the urogram, usually appears as a large sharply defined filling defect in the dome with irregularity of neighboring mucosa simulating carcinoma of the bladder.

Bullous edema is an inflammatory change of the bladder mucosa seen in acute cystitis. The mucosa appears irregularly thickened, inflammed, and rigid, and it is covered by multiple small, rounded, cyst-like elevations resulting from obstruction of the lymphatic channels and submucous effusion. Because of the difficulty in differentiation from tumor we are presenting several examples of this entity. It is important to be aware of the possibility of "catheter edema," although tumor must still be considered in the differential diagnosis.

FIG. 1. Case I. Excretory urogram showing defect (arrow).

REPORT OF CASES

CASE I (Fig. 1). A seventy year old white man was admitted to the Urology Service with signs and symptoms of prostatism. The patient was in acute urinary retention and was catheterized on the day of admission. The Foley catheter was in place for four days prior to excretory urography. The roentgen report stated, "In the region of the right ureteropelvic junction, on the superior margin of the bladder, there is a definite semilunar shaped filling defect associated with irregularity of the bladder wall. The findings here are consistent with a neoplasm of the bladder and should be further studied."

At the time of transurethral resection of the prostate, cystoscopy revealed bullous edema in this area with no evidence of tumor.

CASE II (Fig. 2). A sixty-eight year old male Negro was admitted to the Urology Service with the diagnosis of benign prostatic hypertrophy. The patient had had an indwelling Foley catheter for three weeks prior to excretory urography. The roentgen report stated that the "bladder has an irregular outline, especially on the left side, suggesting the

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FIG. 2. Case II. Excretory urogram showing defect (arrow).

possibility of an intrinsic neoplasm of the bladder."

Cystoscopic examination revealed bullous edema, most marked on the posterior superior surface of the bladder on the left.

CASE III (Fig. 3). A seventy year old white male was admitted to the Urology Service in acute urinary retention. He was placed on Foley catheter drainage for five days prior to excretory urography. Again the defect was noted and the possibility of bladder tumor was suggested.

Cystoscopy revealed the area to contain bullous edema, and there was no evidence of bladder tumor.

CASE IV (Fig. 4). A sixty-nine year old male Negro was admitted to the Urology Service in urinary retention with a Foley catheter which had been in place for three weeks. Excretory urography revealed poor filling on the left side of the bladder suggestive of tumor.

FIG. 3. Case III. Excretory urogram showing defect (arrow).

FIG. 4. Case IV. Excretory urogram showing defect (arrow).

This area was found to be edematous at the time of cystoscopy and no evidence of bladder tumor was seen.

CASE V (Fig. 5). A seventy year old white male was admitted to the Urology Service with signs and symptoms of outlet obstruction. Because of a large amount of residual urine, a Foley catheter was put in place and was present for four days prior to excretory urography. The roentgen report of the pyelogram revealed "marked irregularity of left uretero-vesical junction which could represent a bladder tumor."

Cystoscopy demonstrated edema surrounding both ureteral orifices, most marked on the left. No evidence of bladder tumor was found.

COMMENT

All excretory urograms were made prior

FIG. 5. Case V. Excretory urogram showing defects at both ureteral orifices (arrows).

to transurethral resection. The 5 cases described above showed the characteristic changes of bullous edema. All of the cases presented a similar roentgen appearance, *i.e.*, a sharply defined filling defect in the superolateral portion of the bladder dome, the area where the catheter bag most frequently rests. The findings were constant and readily seen on all urograms in which the bladder was filled with contrast material.

Each of the patients reported was admitted to the urologic service with a diagnosis of benign prostatic hypertrophy. All had been on Foley catheter drainage for lengths of time varying from four days to one month. They were cystoscoped prior to transurethral resection of the prostate gland and in all cases the described filling defects and prominent folds were caused by bullous edema. The mucosa was seen to be pale and swollen and it "pitted" readily when the loop of the resectoscope was pressed into the area. In no instance was there a suggestion of tumor and biopsy was not deemed necessary by the surgeon. None of the patients had clinical evidence by history or urinalysis of either gross or microscopic hematuria prior to catheterization.

SUMMARY

Several cases of bullous edema of the bladder caused by continuous contact with a Foley catheter are reported. The importance of this finding is noted in view of the relative frequency of its occurrence and of its resemblance to bladder tumor.

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GYNECOGRAPHY

TECHNIQUE AND INTERPRETATION

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ROENTGENOGRAPHIC visualization of the structures in the female pelvis by intraperitoneal gas contrast study was first described by Goetze,[7,8] in 1918. Utilization of this technique is occasionally reported in the literature.[1,2,5,6,9,14,15,16,17] However, few radiologists and other clinicians resort to this technique in the diagnostic evaluation of the pelvis or abdomen, mainly because of fear of gas embolism. This fear appears quite unwarranted with choice of proper gas and technique. Indication for this procedure, preparation of the patient, equipment, technique, problems and complications, as well as interpretation will be discussed.

Initially, gynecography was done at the Clinical Center of the National Institutes of Health as part of the evaluation of patients with Stein-Leventhal syndrome[18]. Subsequently, other abdominal and pelvic diagnostic problems in patients ranging from two to seventy-two years of age have been evaluated by this procedure. This very informative test is simple and safe; it may be performed on outpatients, even women weighing 300 pounds.

PREPARATION AND POSITIONING OF THE PATIENT

Careful preparation is the cornerstone of a successful study. A cathartic, preferably castor oil, is given on the day preceding the examination. In the morning the patient has nothing by mouth. A cleansing enema is given one to two hours before reporting to the radiology department. The patient is instructed to empty her bladder "completely" just before the procedure itself is begun. If adequate preparation is questioned, a scout roentgenogram of the lower abdomen may demonstrate a distended bladder or a scybalous rectosigmoid colon.

The patient is placed supine on a standard roentgenographic tilt table that has a footboard and shoulder rest. Her feet are secured to the board by a boot and strap arrangement of the type used in myelographic procedures. Improvisation with broad adhesive tape as used in leg traction will, however, suffice (Fig. 1, *A*, *B*, and *C*).

MATERIALS

To properly perform gynecography, the following materials are necessary:

- Stand
- Gloves
- Sterile peritoneal puncture tray
  - Sponges and spongestick for skin preparation
  - Towels (4)
  - Syringes, ml. (2)
  - Needles, 22 gauge, 1 inch (2)
    20 gauge, 2 inch spinal (1)
    20 gauge, 3½ inch spinal (2)
    20 gauge, 6 inch spinal (1)
  - Tubing, latex, two sections, each approximately 2 feet long
  - Glass tube connector, Y-shaped
  - One way stop cock, syringe adapter
  - Small hemostat (1)
  - Medicine cups (2)
- Xylocaine, 1 per cent
- Sterile saline or water
- Antiseptic solution or tincture
- Tank of nitrous oxide with attached flowmeter, as used in anesthesia apparatus (separate nitrous oxide flowmeters are available from anesthesia equipment manufacturers)

The two sections of tubing are connected by the Y-tube glass connector; one open end of the rubber tubing is attached to the flowmeter of the nitrous oxide tank. The stop cock is connected to the other open end

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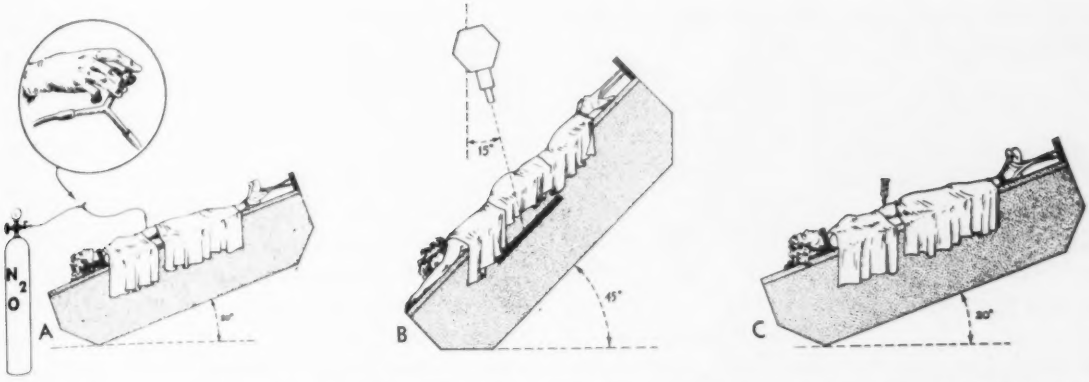

FIG. 1. In sequence these drawings demonstrate (*A*) positioning of the patient during establishment of pneumoperitoneum (note glass connector valve arrangement), (*B*) positioning of patient and equipment during roentgenography, and (*C*) positioning during removal of the gas from the peritoneal cavity (note gas bubbling through the fluid in the syringe).

of the tubing. This entire system is carefully checked for leaks by occlusion of the free end of the Y-tube glass connector.

ESTABLISHMENT OF PNEUMOPERITONEUM

Following preparation of the skin with antiseptic solution, the abdomen is draped with towels, leaving a small portion of the para-umbilical area exposed. Local anesthesia, 2–4 cm. to the left and below the umbilicus, precedes the peritoneal puncture which is made with a spinal needle of proper length. In patients having left sided scars or masses, a corresponding puncture site is chosen on the right; right sided punctures have been found equally sucessful. A 2 inch needle is used on the average patient. Obese women generally require a needle $3\frac{1}{2}$ inches long; the 6 inch needle has been used only twice, each time on a patient weighing over 300 pounds. The needle is slowly advanced until it enters the peritoneal cavity. In most instances it is not difficult to realize when this has occurred. Two distinct "pops" are felt as the needle is advanced: the first may be related to puncture of the fascial plane anterior to the preperitoneal fat, and the second signals entrance into the peritoneal cavity itself. The obturator is removed for inadvertent blood vessel penetration check by syringe (which we have never observed). If no blood is aspirated, the needle is secured by a hemostat resting on the abdominal wall. The table is then tilted 20–25 degrees, headdown.

The stopcock is connected to the spinal needle and opened to the gas line. Flow rate of nitrous oxide is set at 600 ml./min. Timing of gas flow is begun when the radiologist closes the system from tank to patient by occluding the open limb of the Y-tube glass connector with his gloved finger. After a few seconds and several times thereafter during the filling period, pressure is checked by momentary lifting of the finger from the open Y-limb. Only a gentle hissing sound should be heard. A louder noise by the escaping gas indicates increased pressure in the system and mandates repositioning of the needle before proceeding. Two to three thousand milliliters of nitrous oxide are introduced into the peritoneal cavity over three to five minutes. The exact volume used is determined by the patient's size and by the tone of her abdominal wall.

The patient is tilted head-down during filling to prevent abdominal discomfort and shoulder pain resulting from subdiaphragmatic gas accumulation. With this precaution the patient should experience nothing more than increasing abdominal "fullness." If complaints of pain do occur, one can be fairly certain that the needle is improperly positioned and that repeat

peritoneal puncture is indicated before proceeding.

ROENTGENOGRAPHIC TECHNIQUE AND FOLLOW-UP

When the patient has no pain during filling, the total intended volume of gas is delivered, the stopcock closed, the needle removed, and the patient turned into the prone position. Table tilt is increased to 45 degrees head-down. The roentgen-ray tube is angled 15 degrees from the vertical towards the feet, resulting in a 30 degree pelvic inlet projection. Gynecograms are obtained in the posteroanterior and both oblique projections with the central ray entering 3 to 5 cm. below the tip of the sacrum. When the posteroanterior gynecogram furnishes the desired information, the other exposures can be omitted. Occasionally, special problems require other angles and projections.

The technical factors used are: 40 inches target film distance, conventional film and par screens, Bucky grid (8:1 ratio), either phototiming at S-1 selector or 30 to 60 mas., 60–80 kv.

After satisfactory (wet reading) gynecograms have been obtained, the patient is turned back into the supine position and the table tilt is reduced to 20 to 25 degrees. Peritoneal puncture is again performed through the same anesthetized site, a syringe containing 1 ml. of sterile saline or water is attached and the plunger is removed. When bubbling of the escaping nitrous oxide has ceased, the needle is removed and the patient helped from the table. We have our patients lie recumbent for two to three hours before returning home; but this precaution is not essential. Occasionally, a patient will complain of shoulder ache on sitting; this disappears by morning and has not required the use of analgesics. In general, no follow-up observation is necessary.

The above described technique has been found safe and well tolerated in patients ranging from two to seventy-two years of age.

COMPLICATIONS AND PROBLEMS

Cautious, slow and gentle manipulation of the needle is a good way to prevent complications. No serious complications were encountered. Respiratory embarrassment was never a complaint. In one instance gas was introduced into the small bowel (Fig. 7), but the patient experienced no discomfort of any kind and remained asymptomatic over the succeeding days. Physical examination showed promptly subsiding distention. This patient was known to have had suppurative peritonitis with surgical drainage two years earlier, and widespread adhesions were the likely cause of small bowel puncture.

All our problems, minimal and few as they are, derive from improper needle placement. In the very obese or very muscular patient, the previously described sensation of "popping," when resistance is overcome, is not clearly discernible, and preperitoneal needle positioning may result. In the very thin patient inadvertent puncture of the retroperitoneal space is possible. These extraperitoneal insufflations cannot be detected reliably by manometric measurement or by noting of flow rate and resistance with the injection of a few milliliters of sterile fluid. Auscultation of the abdomen during insufflation is sometimes helpful, particularly in the small or normal sized patient; a distinctive hissing may be heard when gas freely enters the peritoneal cavity. The most reliable criterion for improper needle placement and extraperitoneal insufflation is the patient's complaint of discomfort. This takes the form of radiating pain toward the flanks or epigastrium, usually within a few seconds of the onset of gas flow. For the occasional stoic patient even this criterion fails. In such a case, and, in general, if there is some question of the exact needle position, an anteroposterior scout roentgenogram of the abdomen is taken. With perfect intraperitoneal injection, gas sharply delineates the peritoneal cavity inferiorly and laterally (Fig. 2). Preperitoneal extravasation of gas is diagnosed by the mottling that is charac-

teristic of interstitial emphysema; in the pelvis this mottling is noted surrounding the bladder (Fig. 3, 4 and 5). With retroperitoneal infiltration, linear dissection of gas is found along the psoas shadows (Fig. 6).

Extravasation of gas outside the peritoneal cavity occurs infrequently and has caused no complications. Slight discomfort is induced but absorption of small amounts of nitrous oxide from interstitial tissues is complete within about two hours. When gas is not removed from the peritoneal cavity proper after the gynecograms have been taken, complete absorption requires up to forty-eight hours. In this event, shoulder ache upon assumption of the upright position is a common complaint. The advantageous use of nitrous oxide merits brief mention. Teschendorf[20] has shown, and it is well known, that carbon dioxide and nitrous oxide are "safe" gases that are quickly diffused and absorbed. Resorption time of these two gases is approximately twenty times faster than that of oxygen.

Fig. 2. (29144). Anteroposterior scout roentgenogram demonstrates sharp delineation of the peritoneal cavity laterally and inferiorly after proper gas insufflation. (Figure 1*A* shows the position of the patient and the table; the central ray enters perpendicularly to the table).

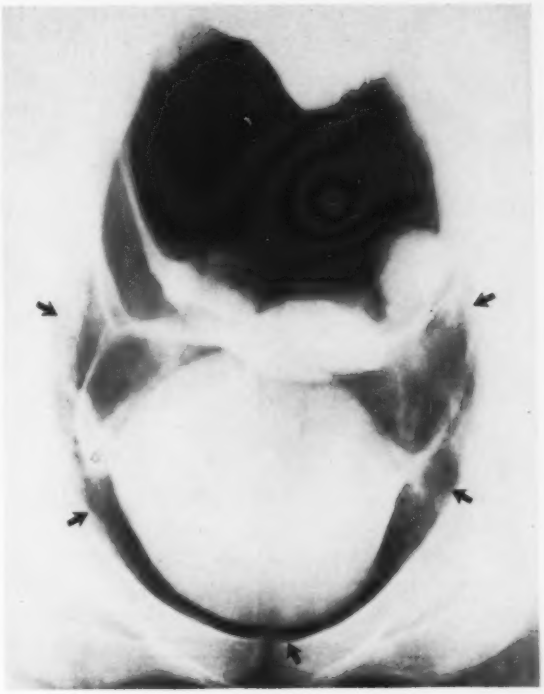

Fig. 3. (30338). Gynecogram shows interstitial emphysema due to preperitoneal extravasation of gas (arrows). Note the clear delineation of the urinary bladder by the surrounding gas. Repositioning of the needle resulted in good intraperitoneal gas contrast with effective visualization of the pelvic organs.

The ratio is even greater when compared to air. Carbon dioxide and nitrous oxide are nontoxic. Although carbon dioxide is absorbed slightly faster than nitrous oxide, the latter gas is less irritating and for this reason is preferred in our department.

Interpretation of Gynecograms

Well performed gynecography reveals the pelvic organs with unusual clarity. Roentgen findings in an anatomically normal pelvis (Case 1) are shown in Figures 8 and 9.

Case 1 (28882). A thirty-two year old subject with Cushing's syndrome had normal menses, no abnormal findings on pelvic examination and a normal endometrial biopsy. At laparotomy for bilateral adrenalectomy, the ovaries and uterus were unremarkable. The pathologic diagnosis was adrenocortical hyperplasia. Ovarian biopsies were normal.

Anteriorly, the smooth shadow of the col-

FIG. 4. (28713). Gynecogram shows partial preperitoneal extravasation (black arrows) in a patient with normal ovaries and uterus. Note excellent visualization of the round ligaments (white arrows).

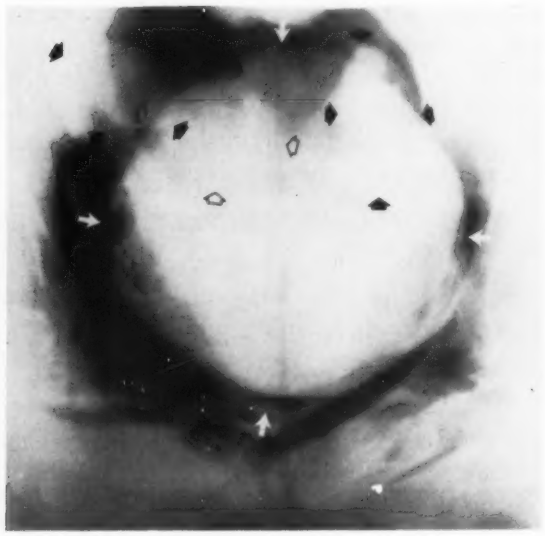

FIG. 5. (24591). Gynecogram shows extensive preperitoneal gas extravasation. Note the faint visualization of the enlarged ovaries (black arrows) and of the uterine fundus (outlined arrows) and the clear delineation of the bladder (white arrows).

FIG. 6. (29595). Gynecogram shows retroperitoneal gas extravasation causing linear dissection of gas along the psoas muscles (arrows).

lapsed bladder was readily recognized on the gynecogram. Both adnexal regions were smooth and free of masses. The uterine fundus was seen in the midpelvis, with its density superimposed on that of the cervix. Bilaterally, the curvilinear shadows of the round ligaments branched away from the broad ligaments anteriorly. The fallopian tubes with their thickened fimbriated ends were seen intimately associated with the ovarian shadows in the superior lateral aspect of the broad ligaments. The ovaries themselves were posterolateral to the uterine fundus; their precise location relative to the fundus was variable and changed with shifting position of the patient. The collapsed rectosigmoid reflection extended posteriorly and along the pelvic rim to the left and upward into the abdomen.

The size of the pelvic organs is readily determined from the measurements made on the posteroanterior and both oblique gynecograms. The width of the uterine fundus is conveniently measured from isthmus to isthmus. The fundus thickness is determined perpendicular to this interisthmus line on the posteroanterior film. The ovarian length is the longest dimension on the

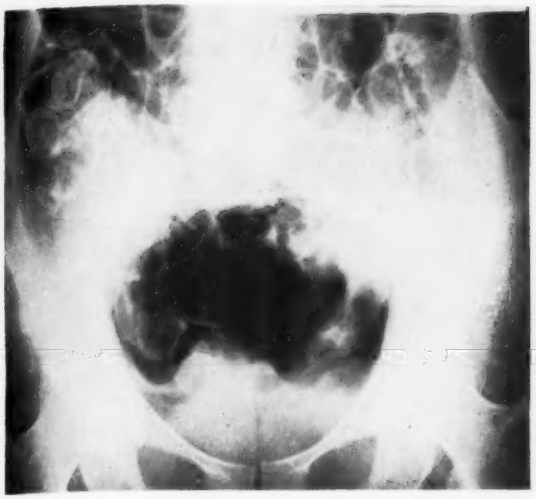

Fig. 7. (27055). Gynecogram shows accidentally introduced gas in the small bowel of a diabetic patient who had had suppurative peritonitis and surgical drainage two years earlier. This caused no symptoms or sequelae.

posteroanterior or either oblique gynecogram. The measurement of the greater dimension at right angle to this long axis yields the ovarian width; the lesser perpendicular dimension is the ovarian thickness. All measurements should be corrected for distortion by decreasing the numbers by 20 per cent on patients of average girth and by 25 per cent on obese patients.

In cases where gynecographic measurements were checked at laparotomy, correlation was good and the greatest discrepancy only once exceeded 1.0 cm. In most instances, roentgenographic measurements varied by no more than a few millimeters from the dimensions recorded at operation.

The pelvic viscera of normal females of child bearing age vary considerably in size. Standard textbooks of anatomy[13] and obstetrics[4] record normal ovaries measuring 2.5 to 5 cm. in length, 1.5 to 3 cm. in width and 0.7 to 1.6 cm. in thickness. An even greater range is given for dimensions of the normal nongravid uterus. Despite this variation in size of the viscera themselves, the *ratio* of the uterine fundus width to the ovarian length is consistently greater than 1.0 cm. and usually greater than 1.3 cm. in the normal pelvis.

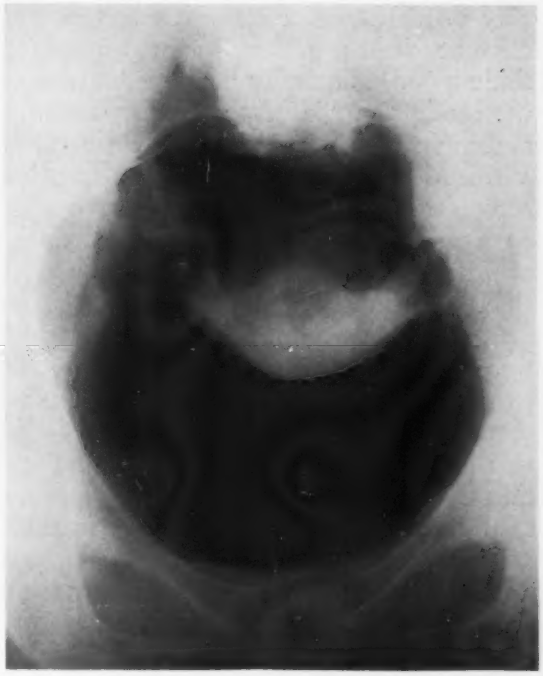

Fig. 8. Case 1 (28882). Normal posteroanterior gynecogram (see Fig. 1*B*).

In this hospital, gynecography has been used frequently in the evaluation of patients with the polycystic ovary syndrome. This entity, first described by Stein and Leventhal[18] in 1935, was recently reviewed by Stein.[19] Pertinent features are the his-

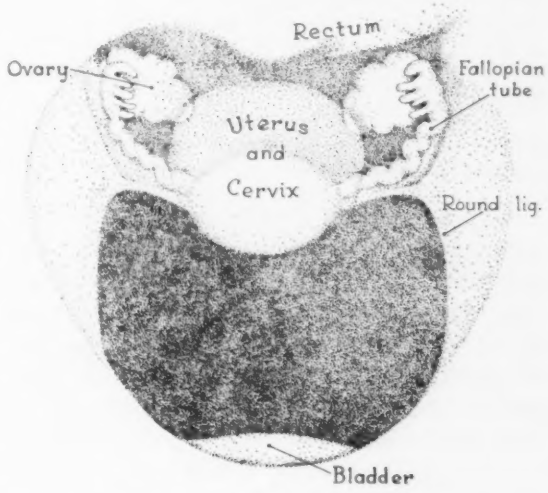

Fig. 9. Case 1 (28882). An illustrative drawing of Figure 8 is presented since copying and recopying of the original gynecogram tends to obscure detail.

tory of menstrual irregularities (especially oligomenorrhea or amenorrhea) and the finding of bilaterally enlarged ovaries, uterine hypoplasia in 75 per cent and hirsutism in about 50 per cent of the cases. Virilization is absent. Stein and Leventhal recognized at the outset that the ovarian enlargement, crucial to the diagnosis, could not be established in about half of their patients by physical examination alone. Using gynecography, however, they were able to demonstrate unequivocal ovarian enlargement in all cases. Gynecograms also delineated the typically small uterine fundus. These authors called attention as well to the usefulness of the ratio of the ovarian to the uterine size. In their experience the normal ovary was one-fourth to one-half the size of the uterine fundus, whereas the polycystic ovary was at least three-fourths as large as the uterine shadow.

Gynecograms of a typical case of the Stein-Leventhal syndrome[18] (Case II) are shown in Figure 10, *A*, *B* and *C*. Figure 10*D* shows another patient with the same syndrome.

Case II (22002). An eighteen year old girl, complaining of hirsutism, oligomenorrhea and obesity, had a small uterus but no suggestion of ovarian enlargement on physical examination. At laparotomy the ovaries were found to be large and cystic. The microscopic findings were compatible with those described by Stein and Leventhal (cf. Stein review). Gynecographic demonstration of the ovarian enlargement relative to the small uterine fundus was the decisive factor leading to exploration.

Ovarian enlargement in the polycystic ovary syndrome is described as bilaterally symmetric (Stein review). We have confirmed this finding in all but one of our patients with this entity. This patient (Case III) had bilateral ovarian enlargement but the enlargement was asymmetric (Fig. 11). Gross and microscopic findings were typical of those described by Stein and Leventhal.

Case III (27726). A twenty year old student nurse had menarche at the age of eleven, oligomenorrhea from the age of eleven to sixteen and menorrhagia for the next two years, followed by progressive oligomenorrhea. At the time of her admission she had been amenorrheic for seven months. Severe acne and moderate hirsutism were apparent on physical examination but her habitus was female, her breasts well developed and her clitoris normal in size. The right ovary was enlarged, questionably cystic and very tender to palpation. The left ovary was felt to be somewhat smaller but was also tender. The twenty-four hour excretion of 17 ketosteroids was normal. Surgical exploration revealed a right ovary measuring 5.0×3.5×2.8 cm. and a left ovary measuring 3.7×2.9×2.4 cm. Bilateral wedge resections were performed. Microscopically, the ovarian wedges showed an extremely thick and fibrous tunica, subcapsular cysts, no corpora lutea and only one corpus albicans.

Ovarian enlargement of the Stein-Leventhal syndrome is not associated with adnexal abnormalities, attachment of the ovaries to the adnexa, or abnormal configuration of the broad ligaments (Fig. 10, *A–D*; and 11). One patient (Case IV) with suspected polycystic ovaries was found at gynecography to have symmetric bilateral ovarian enlargement, but the ovaries were attached to irregular and thickened adnexal regions (Fig. 12).

Case IV (18266). A twenty-eight year old Negro woman had oligomenorrhea, infertility and a history of a vaginal discharge ten years earlier for which she had been treated with penicillin. The uterus and adnexal areas were normal to palpation. She was not hirsute. Serology was negative. Urinalysis and renal function were normal. The preoperative diagnosis was probable Stein-Leventhal syndrome. At surgery she was found to have chronic pelvic inflammatory disease in addition to polycystic ovaries. Hysterosalpingograms made after surgery demonstrated patency of the right and obstruction of the left fallopian tube; numerous diverticula arose from each fallopian tube.

The usefulness of gynecography is well demonstrated in another patient (Case V) suspected of having the polycystic ovary syndrome (Fig. 13, *A*, *B* and *C*).

Case V (29922). This twenty-seven year old white woman had oligomenorrhea, infertility,

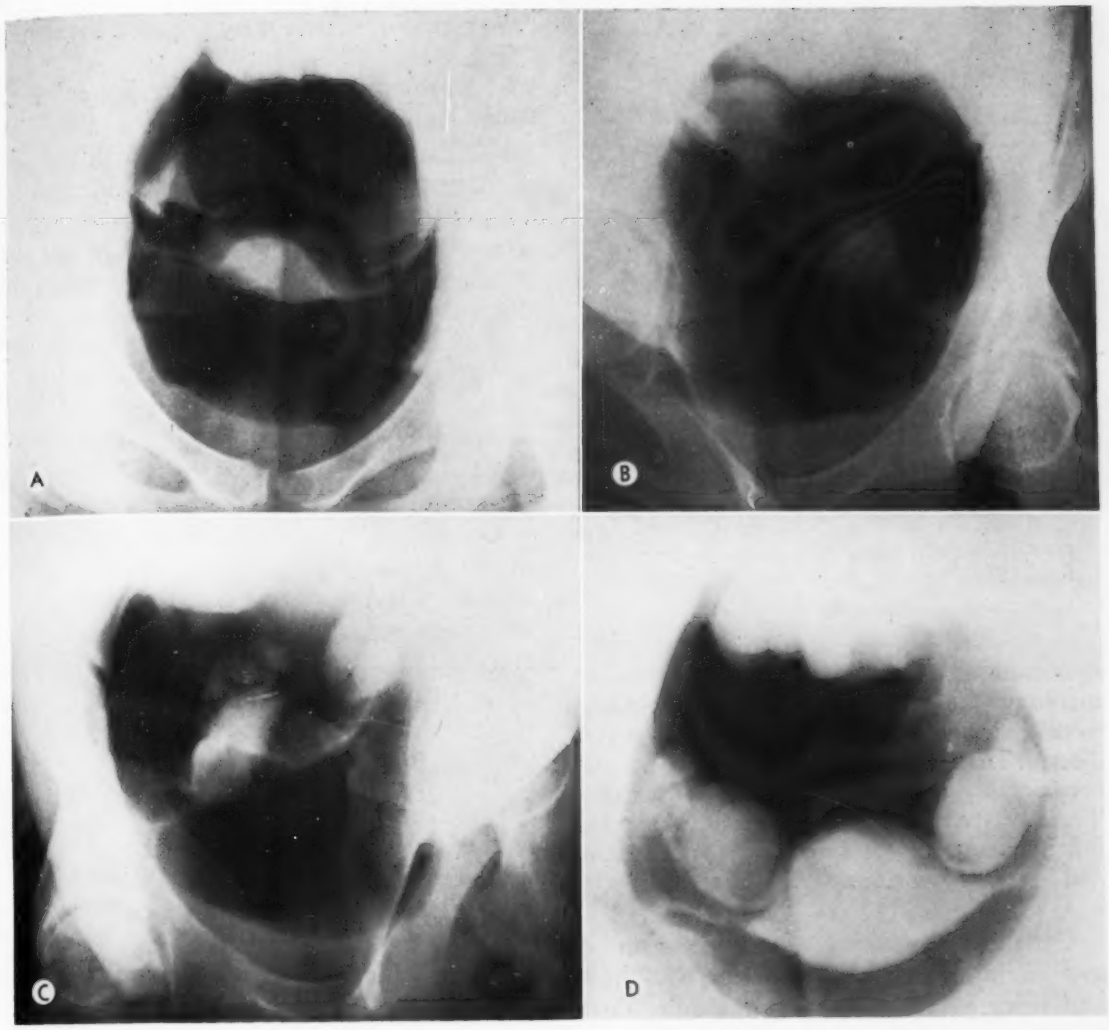

Fig. 10. Case 11 (22002). Stein-Leventhal syndrome. Posteroanterior (*A*), right anterior oblique (*B*) and left anterior oblique (*C*) gynecograms demonstrate large, thick ovaries and a relatively small uterine fundus. Note the smooth bladder shadow anteriorly, the collapsed rectosigmoid colon posteriorly, the cervical shadow superimposed upon the fundus shadow and the sharp delineation of the fallopian tubes. In the oblique studies (*B* and *C*), note the shift of the ovaries with the shift of the patient. (*D*) (31161). Gynecogram shows excellent visualization of the enlarged ovaries and of the uterus in another patient with the Stein-Leventhal syndrome. Note the fimbriated ends of the fallopian tubes.

and hirsutism. There was no history of venereal disease or abdominal surgery. Pelvic examination was unsatisfactory; the ovaries were not readily palpable but considered to be somewhat large. One senior observer felt "right adnexal fullness." She was sent to the radiology department with the diagnosis of Stein-Leventhal syndrome.

Gynecograms (Fig. 13, *A*, *B* and *C*) demonstrated a large pelvic mass, adherent to the right adnexa, widened and tortuous uterine ligaments bilaterally (fallopian tubes), a fixed, lobulated, slightly enlarged left ovary and a uterus that was small but probably within normal limits in size. A hysterosalpingogram (Fig. 14) showed the changes of bilateral old tubal inflammatory disease. At laparotomy bilateral chronic pelvic inflammatory disease was evident. Peritoneal adhesions, thickened, fixed and tortuous fallopian tubes, a right ovarian tumor and a cystic, perhaps slightly enlarged, left ovary were noted during dissection. The right ovarian mass measured 6.5×4×3 cm.; histologic sections showed papillary

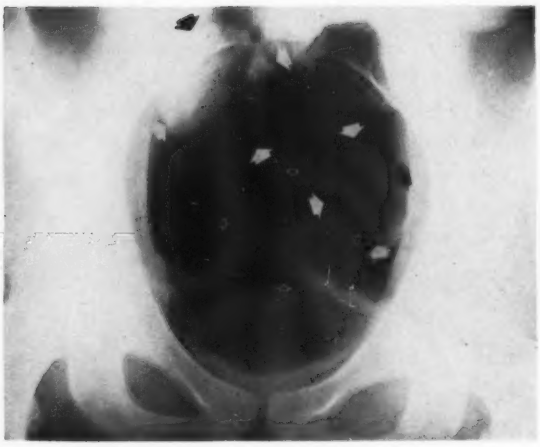

FIG. 11. Case III (27726). Gynecogram shows the ovaries of this patient with the Stein-Leventhal syndrome to be asymmetrically enlarged (large arrows). The small arrows delineate the uterine fundus.

cystadenoma. Multiple cysts were noted on incising the left ovary; the microscopic findings were considered compatible with polycystic disease. The roentgenologic findings contributed considerably to the management of this case. There was no clinical evidence for the pelvic inflammatory disease shown by gynecography and hysterosalpingography.

Generally speaking, interpretation of gynecograms done on patients who have had prior pelvic surgery is difficult. Postoperative adhesions often prevent the free entrance of gas into the pelvis and all but one of the gynecograms performed on such patients were technically below par.

Some difficulty is encountered as well in the reading of gynecograms where there is borderline enlargement of the ovaries in the presence of a relatively small uterus (Fig. 15); in such cases we recommend culdoscopy.

A gynecogram (Fig. 16) made of a fifteen year old girl (31339) who was suspected of having ovarian agenesis showed a very small cervix and very small and rudimentary genitalia, consisting of mere soft tissue ridges, extending toward both adnexal regions, and ending in oval shaped thickenings. This finding was not established by the rather unsatisfactory physical examination.

Figure 17 demonstrates ovarian enlargement, more marked on the left, and uterine enlargement in a thirty-two month old patient (31872). This precocious girl began to menstruate at the age of eleven months. Repeated rectal examinations were not revealing. Ovarian biopsies showed multiple follicular cysts.

Carcinoma of the cervix demonstrated by gynecography has been described previously.[1] Cervical, broad ligamental and adnexal changes due to tumor invasion were observed in patients studied primarily for measurements of "normal" ovaries.

Occasionally, other primary and metastatic tumors, particularly of the lower abdomen and pelvis, were successfully evaluated by gas contrast studies. Consideration of these cases is, however, beyond the scope of this paper.

FIG. 12. Case IV (18266). Gynecogram shows chronic pelvic inflammatory disease in this patient with the Stein-Leventhal syndrome. Note fixation of enlarged ovaries by adnexal adhesions (large arrows) and the small uterus and cervix (small arrows).

FIG. 13. Case v (29922). (*A*, *B* and *C*) This patient had papillary cystadenoma of the right ovary, Stein-Leventhal syndrome, and chronic pelvic inflammatory disease. Note the large fixed right pelvic mass and the enlarged fixed left ovary (broad arrows), thickened ligaments (tubes) bilaterally (slender arrows), and the uterus and cervix.

DISCUSSION

Gynecography is a simple but very useful technique in diagnosis. Using nitrous oxide as a contrast agent, the procedure carries a negligible risk of mortality and no cases of fatal gas emboli have been reported with this highly water soluble gas. Morbidity has been rare, transitory, and without sequel.

Excellent visualization of the pelvic

FIG. 14. Case v (29922). Hysterosalpingogram shows evidence of chronic inflammatory disease (arrows). Peritoneal spillage of contrast material on the right side was observed on a subsequent roentgenogram.

structures was achieved in all patients who had not undergone previous abdominal or pelvic surgery. In all but one of those who had had antecedent surgery, the gynecograms were still adequate for evaluation of ovarian and uterine size and configuration. The accuracy of pelvic organ measurement by this method was repeatedly confirmed at laparotomy.

FIG. 15. (28854). Gynecogram shows borderline enlargement (particularly thickening) of the ovaries. In such instances culdoscopy is suggested. Note a loop of sigmoid colon overlying the left ovary.

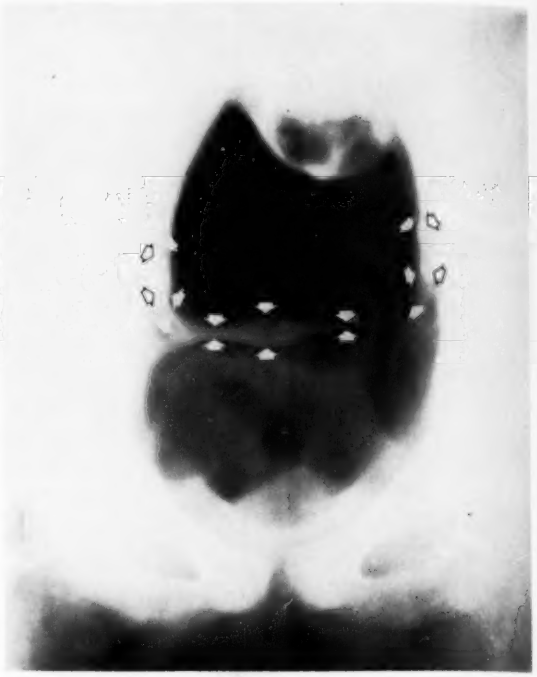

FIG. 16. (31339). Gynecogram shows ovarian agenesis in a fifteen year old girl. Note the rudimentary genital structures (arrows).

Gynecography is particularly useful in evaluating the pelvis of extremely obese or very young patients. In the former, physical examination, even under anesthesia, is often unrewarding. In children, the risk of general anesthesia is probably greater than risk of pneumoperitoneum—and the physical examination less informative than the roentgenogram. Gynecograms provide objective confirmation of positive physical findings and, notably in the polycystic ovary syndrome, will unearth ovarian enlargement missed by the clinician. We have been impressed as well with the opportunity to diagnose chronic pelvic inflammatory disease by roentgenography where there had been little or no clinical evidence to suggest such an entity.

Gynecography has been superseded in at least one large clinic[11] by culdoscopy, an

endoscopic procedure affording direct visualization of the pelvic contents. Even in experienced hands, however, the use of the culdoscope carries a certain morbidity.[12] It is our impression that this morbidity, however small, is greater than that associated with nitrous oxide pneumoperitoneum and hence we advocate gynecography as the initial screening procedure. This is particularly true in situations where previous abdominal surgery or pelvic inflammation has increased the risk of a culdoscopic accident.

We have no intention of minimizing the role of culdoscopy. Rather, we wish to emphasize the desirability of doing first that procedure, of least risk to the patient, which can still furnish sufficient indication for laparotomy.

SUMMARY

1. Visualization of the pelvic organs by gas contrast roentgenography (gynecography) is a useful diagnostic technique.

2. The method is simple. Instruments and equipment should be readily available in the radiology and supply departments of any hospital.

3. Using nitrous oxide gas, no fatalities have been reported. Morbidity is small in incidence and trivial in extent.

4. The method of patient preparation, equipment operation, roentgenographic technique and film interpretation, as well as the problems encountered, are described.

5. Several clinical problems are presented and gynecograms are illustrated.

6. The relative roles of gynecography and culdoscopy are briefly discussed.

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Fig. 17. (31872). Gynecogram shows ovarian enlargement in a thirty-two month old girl with precocious development and menarche at the age of eleven months. Note the ovaries (thick arrows) and uterus (outlined arrows) superimposed upon the partially filled urinary bladder.

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Vol. 86, No. 5

BREAST ROENTGENOLOGY*

HISTORICAL REVIEW

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A REVIEW of the historical development of breast roentgenology is presented so that the experienced roentgenologist might obtain a better orientation to a roentgenographic discipline of great value in the detection of early cancer as well as other diseases of the breast.

In this country, roentgenology of the breast gained recognition in the 1950's as a useful diagnostic procedure. Many radiologists, however, still classified the examination as a special procedure, if not a technical ordeal, and many surgeons and practitioners were only vaguely aware that such an examination could be performed. Requests were few and when examinations were performed they were done often in a perfunctory manner. Most radiologists were unfamiliar with the appearance of normal breasts and, when presented with a diseased breast, were hesitant to interpret even the most clear-cut evidence of disease.

This brief review will point out some of the notable achievements as well as the many serious failures of early investigations. It will also serve to indicate areas still needing investigation, so that others may be encouraged to exploit this field to its full potentialities. Those interested in an exhaustive bibliography are urged to consult the comprehensive review published by Professor J. Von Ronnen[35] of the University of Leyden.

In 1913, a German surgeon, Salomon,[30] was the first to take roentgen-ray "pictures" of the breast. He used excised breasts, since his primary concern was to study the extent and mode of spread of mammary cancer in order that a more adequate biopsy specimen could be removed at the time of surgery. His observations of 3,000 mastectomy specimens correlated the roentgen findings with the gross and microscopic anatomy of the tumors. It is apparent that Salomon did not take full advantage of the implied diagnostic applications of his findings. However, the case histories and the descriptions and actual reproductions of the roentgenograms contain a wealth of interesting information which had considerable impact on subsequent investigators.

Salomon's most important observation was the recognition that a roentgenogram gave a true picture of the margins and extent of a tumor. He also recognized the differences in roentgen appearance of the most common forms of mammary cancer. He clearly differentiated the scirrhous or infiltrating type from the circumscribed or nodular form. Another common type, the duct carcinoma with punctate calcifications, was not recognized as a separate entity, although later duly categorized by another radiologist, Leborgne.[25] The characteristic roentgen appearance of these calcifications, however, is demonstrated in one of Salomon's illustrations. Since this appearance later acquired real diagnostic significance, the original caption is worth paraphrasing: at the center of a rather large infiltrating tumor there were many tiny calcific densities which stood out against the background on the tumor itself. Microscopic study traced the calcific deposits to cystically dilated spaces within a tumor growing, for the most part, intraductally.

In another remarkable case Salomon reports the first "clinically occult" breast cancer found by roentgen examination. In this case the breast had been removed because of a large palpable cyst. Histologic sections taken from the obvious lesion were reported as benign by the pathologist. On

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the roentgenogram of the specimen, two lesions could be clearly identified: a large, rounded, sharply defined cyst, and a tiny, very dense, spiculated tumor with the typical appearance of a scirrhous carcinoma. Unfortunately, the gross specimen in this case was lost or discarded before the roentgenograms became available for study, so no real confirmation could be made. Nevertheless, Salomon expands at some length on the very strong suspicion of malignancy discovered by means of the roentgen-ray picture.

For a number of years following Salomon's paper, there were no references to breast roentgenology in the literature. However, between 1924 and 1930, at least three independent groups were investigating the roentgen diagnosis of breast lesions. Priority is generally assigned to Goyanes, Gentil, and Guedes.[16] These authors present roentgenography of the breast as a new method of diagnosis, especially useful in differentiating inflammatory from neoplastic lesions. They described the positioning required for the examination and the normal image obtained, with its characteristic "planes." Although only a small number (56) of breasts were examined, the report was written to present preliminary findings and to encourage further use and development of the technique.

In addition to the work of Goyanes, Gentil, and Guedes, a "breast clinic" in Leipzig which was actively engaged in taking diagnostic roentgenograms of female breasts was described in Kleinschmidt's textbook.[23] The clinic and the taking of roentgenograms were under the general auspices of Dr. Erwin Payr. Dr. Payr contributed to many areas in surgery throughout a long research career, but he never personally published on the breast project.

Two papers were eventually published as a result of the work done in the Leipzig breast clinic. The later report, by Finsterbusch and Gross,[7] appeared in 1934. It is a case report on the unusual type of calcifications found in what we learned to recognize as secretory disease.[13] The report presents a very careful correlation of roentgen and histologic pathology as well as an attempt at biochemical analysis of the lesions.

The earlier paper by Vogel[34] is a classic in the field. Vogel's work is important for his roentgen classifications of various benign lesions and for his discussion of their differential diagnosis from carcinoma. The criteria used and the care given to his choice of illustrative material are still valid. Though considerably expanded, his observations form the basis of our present day interpretative criteria. Vogel also presented excellent advice on roentgenographic technique.

Although the above contributions were important, a greater impact on American radiologists was made by reports of Warren[36] and Frey and Warren.[8] Their technique—stereoscopic roentgenology—has merit and this area perhaps has not been sufficiently exploited. Warren also contributed some excellent descriptions of the normal gland at rest and during pregnancy. He differentiated the normal from the dysplastic gland and was amazed at the high incidence of the latter in his small series of cases. More important to our present day approach was his imaginative use of the roentgenogram to study breast changes over long intervals of time.

Other outstanding reports during this period were those by Seabold[31–33] and Reimann and Seabold[29] on the roentgen changes in the breast, especially during the normal menstrual cycle. Lockwood,[26,27] Lockwood and Stewart,[28] and Gunsett and Sichel[18] repeated descriptions of diagnostic criteria established by previous workers and defined some areas where inaccuracy could be expected. These difficult areas included microscopic carcinoma and small neoplasms in dense "cystic" breasts.

An often quoted work is a thesis by Espaillat[6] which appeared in Paris in 1933. The text is an attempt to correlate roentgen appearance with symptomatology and microscopic anatomy. It is perhaps the most ambitious attempt at a diagnostic manual in the area of breast roentgenology. This

work is disappointing, however, because of the incomplete analysis of the case material and because of the poor illustrations.

In 1938, Gershon-Cohen and Strickler[15] published an article on roentgenologic examination of the normal breast. They considered that knowledge of the normal breast at all ages and stages of activity was a prerequisite to recognizing pathologic conditions which might arise. The authors acknowledged that improvements in roentgen technique would advance the diagnosis of early neoplastic changes in the breast and endorsed enthusiastic application of the examination.

The literature of this decade (1930's) contains numerous other papers on breast roentgenology originating both in this country and abroad, but it seems to us that very little new information emerged from these publications. The usual pattern was a review of the current literature, followed by a review of various techniques. The diagnostic criteria first reported by the very earliest investigators were repeated and described against the background of experience with a small series of cases. The reports ended with the mention of limitations of the examination and the illustrations, when present, were not informative. It is not surprising at all that a number of people interested in the application of this diagnostic procedure should have become discouraged.

In a way, the initial failure of breast roentgenology is reflected in the first American report by Warren.[36] The main difficulty was the technical quality of the roentgenograms. A great deal of attention was given to positioning and to devices that would spread out the gland. An important diagnostic sign, punctate calcifications found in 30 per cent of all cancer cases, went unrecognized in series after series. The calcific deposits were not visualized on the roentgenograms or, if seen, were confused with artifacts.

A second fault can be traced more directly to Warren's first report,[36] in which he claimed an accuracy of 85 to 95 per cent in a highly selected series of cases. Such a high degree of accuracy impressed subsequent workers who became discouraged when the figures could not be duplicated in unselected series.

Between 1927 and 1931, Domínguez[3,4,5] of Uruguay and Baraldi[1] of Brazil published a series of reports on the use of pneumomammography as a means of obtaining better visualization of lesions in the breast. The technique was adopted in this country by Hicken,[19] who also resorted to injection of the lactiferous ducts and cystic cavities with radiopaque substances. The procedures were apparently intended to be used in cases where visualization could not be obtained by other means.

Raul Leborgne,[24] a student of Domínguez experimented with intraductal injections and he stressed the importance of good contrast which he obtained with very low kv., nonscreen film and good collimation. Use of this technique gave details previously not obtained.

The literature of the 1940's contains numerous references to the dangerous sequelae following the injection of some of these radiopaque substances. The first results were mastitis and abscess; and in isolated cases in which thorotrast was used, cancer developed (Brody and Cullen[2]).

Leborgne[25] in 1951 reported peculiar calcifications in about 30 per cent of mammary cancers, mainly of the intraductal type. Because these tumors occurred more frequently in dysplastic breasts, the sign provided an opportunity for the detection of neoplasms in a type of breast which had previously been the most difficult to examine, both clinically and roentgenographically. Since these tumors grew slowly and spread intraductually and since the patient often had little or no evidence of any abnormality, early discovery of neoplasms by roentgen examinations made clinicians and surgeons realize more fully the diagnostic value of the method.

In 1951 Gros and Sigrist[17] in Strasbourg published the first of many enthusiastic papers on breast roentgenography. They

confirmed the criteria and indications for roentgen examination and their experience with technique and case material parallels that of our own group, especially in the detection of early, clinically occult, unsuspected or asymptomatic tumors.

It is extremely difficult to evaluate our own work and that of our contemporaries at this early date. Our approach to the problem was to base roentgen examination of the breast on a firm anatomic basis. This approach was not new and can be traced as far back as the reports by Salomon, the Leipzig group and Warren. Confusion in terminology existing among surgeons and pathologists in the classification of benign and malignant lesions further complicated progress.

Collaboration with Helen Ingleby, initiated early in 1950, provided the opportunity for a thorough review of the various lesions found in the breast. Opacities seen on roentgenograms were compared with thick whole breast sliced sections of the corresponding specimen and later with the the microscopic material; this approach permitted recognition on the roentgenogram of shadows which had previously appeared unimportant. Observations of abnormal areas on the roentgenogram might call for a search of the surgical specimen for previously unrecognized abnormalities. In more recent years, we were able to correlate roentgen and pathologic findings with certain clinical entities, chiefly adenosis and secretory disease. Use of repeated examinations has been applied to such problems as mammary abscess, pregnancy and lactation and the mode and rate of growth of mammary cancer.[9–14,20–22]

There are still confusing aspects in roentgenology of the breast to be studied and the recent revival of interest in this field is encouraging. Technical improvements of roentgenograms and illustrations, detection of clinically unsuspected carcinomas and the early visualization of lesions for biopsy all have contributed to interest in this field. It is hoped that investigations will continue and that the uncertainties of clinical examination of the breast will be resolved by routine use of this very useful diagnostic procedure.

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THE MOTOR ACTIVITY OF THE ESOPHAGUS IN ASSOCIATION WITH ESOPHAGEAL ATRESIA AND TRACHEOESOPHAGEAL FISTULA*

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PATIENTS who have had surgical repair of esophageal atresia, with or without a tracheoesophageal fistula, often have symptoms as a result of dysphagia, impaction of foreign bodies, and regurgitation with aspiration and subsequent pulmonary infection. These manifestations are usually encountered in the immediate postoperative period, again when solid foods are introduced into the diet, and later in life when solid food is masticated incompletely or swallowed hastily. Cinefluorographic studies of the swallowing act show that narrowing of the lumen of the esophagus at the site of the anastomosis may contribute to the symptoms, but that there is, in addition, a functional disturbance in motor activity of the esophagus of considerable importance.

MATERIAL AND METHOD

Cinefluorographic studies of the esophagus of 15 patients who had a primary anastomosis of the esophagus have been carried out since 1956. In 14 patients, the initial study was performed two to three weeks postoperatively. When subsequent studies were made, these were done at intervals of two months to three years. One patient was examined first at seven years of age and then again two years later. Five patients were examined once, 4 patients were examined twice, and 6 patients were examined three or more times. The patients were examined while prone (left posterior oblique position). A mixture of barium and water was utilized as contrast material. The exposure was made on 16 mm. movie film (Kodak Linagraph Shellburst) at 24 or 32 f/sec. during and after swallowing this material. A 16 mm. Bell & Howell camera, with a 0.95, 25 mm. lens mounted on a 5 inch image intensifier,* was utilized to record the swallowing act.

DISCUSSION

Cinefluorography of normal infants and children reveals a pattern of deglutition comparable to that described as occurring in adults.[2] Deglutition may be divided into three stages. The first stage is voluntary and is concerned with the propulsion of the bolus of liquid or solid material into the pharynx. The presence of the bolus in the pharynx stimulates the reflexes responsible for the second and third stages, which are involuntary. The second stage is characterized by closure of the oral cavity, nasopharynx and larynx; contraction of the pharyngeal constrictors (initiation of the primary peristaltic wave); and relaxation of the cricopharyngeus muscle. The third stage is the continuation of the primary peristaltic wave down the esophagus and the relaxation of the esophagogastric junction. The passage of liquid is as a solid bolus when the patient is supine; when the patient is erect, the liquid precedes the primary peristaltic wave and is held up at the lower end of the esophagus until the wave reaches this area. A succession of swallows of liquid will inhibit the primary peristaltic wave. Secondary waves in the vicinity of the bolus may be initiated by local distention of the esophagus. Reverse peristalsis does not occur in the esophagus,

* Manufactured by North American Philips Company, Inc.

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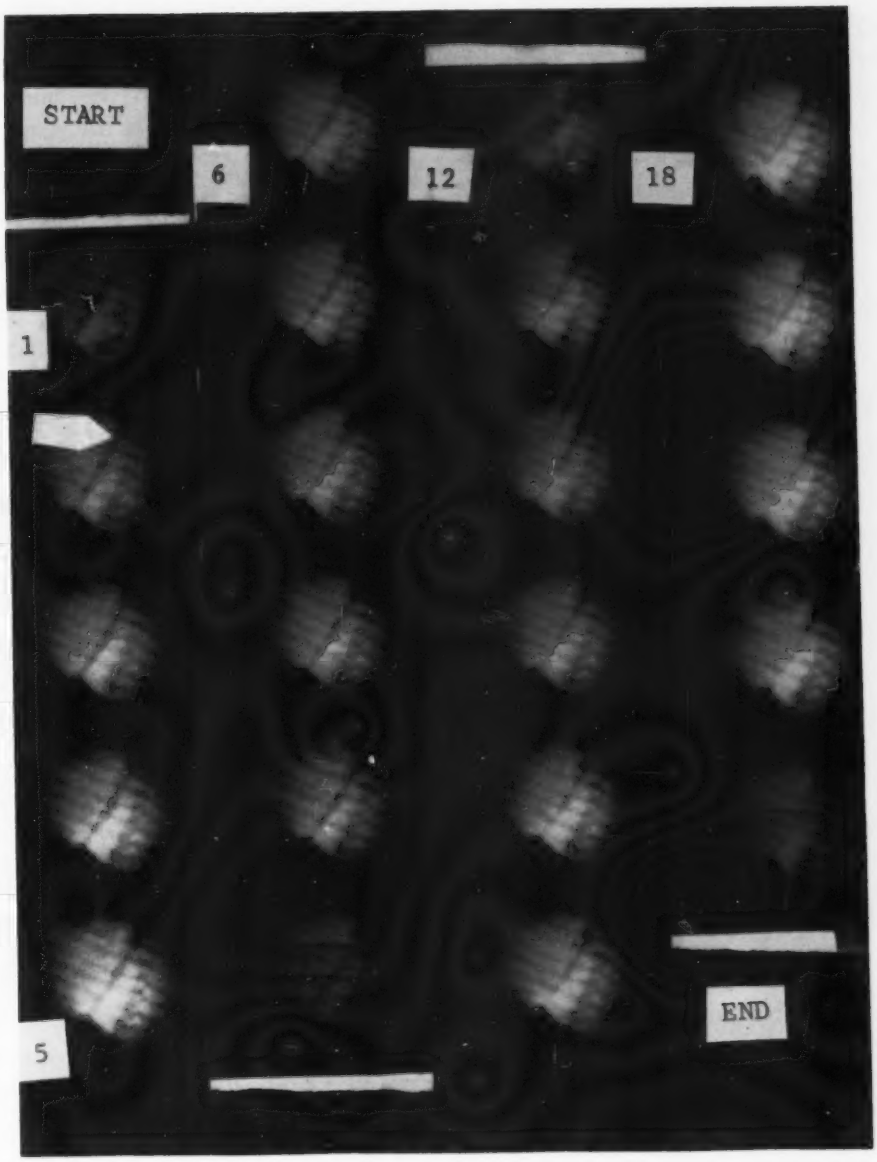

Fig. 1. A selected cine sequence of a patient who had surgical repair of esophageal atresia and tracheoesophageal fistula (left anterior oblique position). The arrow points to the line of anastomosis. Contrast material remains in the esophagus after feeding. The sequence demonstrates contraction of the lower esophagus with retrograde flow of barium into the esophagus above the line of anastomosis.

but a reverse flow of esophageal contents may be caused by nonpropulsive contractions which spread upward.

Patients who have had repair of esophageal atresia, with or without tracheoesophageal fistula, do not show coordinated activity. The primary peristaltic wave is initiated in the pharynx, but does not progress along the length of the esophagus. In many instances, it becomes dissipated above the line of anastomosis so that the contrast material remains in this area until a mass contraction occurs. Then, some of the contrast medium may pass in a retrograde manner into the pharynx, but the bulk usually goes past the line of anastomosis into the lower esophagus. Although the initial impression in these instances

may be that of an organic obstruction at the line of anastomosis, it seems probable that, at least a considerable portion of the obstruction is functional, being dependent upon the disturbed motor activity. Contrast material in the esophagus below the line of anastomosis, if of an amount that does not distend the esophagus, may remain in the esophagus for many minutes until it flows downward. If of an amount to result in distention of the esophagus, secondary contractions may occur that are either localized to the point of maximal distention or are more diffuse and involve most of the lower esophagus. These contractions result in both antegrade and retrograde flow of the contrast material. In several patients, the retrograde flow was sufficient to enter the pharynx and spill over into the larynx and airway. In some patients, the esophagogastric junction was competent; in others, there was free flow from the stomach into the esophagus with crying and subsequent increase in intra-abdominal pressure (Fig. 1).

Not in all patients was the extreme form of the abnormal motor activity apparent, and symptoms were not present in all patients. In all the postoperative patients examined cinefluorographically, however, there was some degree of incoordination of primary peristalsis, particularly below the line of anastomosis. Haight[1] has referred to this abnormality in 5 patients, six to eight and one-half years of age, examined by means of a rapid cassette changer. In one of our patients, seven years of age, the mother stated that the child would regurgitate if he ate too rapidly or masticated insufficiently. He demonstrated deficient motor activity below the line of anastomosis and, at times, retrograde flow in the esophagus but not into the pharynx.

The motor activity in the esophagus in these patients is not unlike that described after the administration of banthine (methantheline), an anticholinergic agent.[3] Thus, one is led to speculate that the abnormality is secondary to interruption of the vagal fibers to the esophagus at the time of surgical intervention. To date, however, it has not been possible for us to correlate the amount of operative dissection with the postoperative activity of the esophagus. We have had the opportunity, moreover, of examining pre-operatively one newborn infant with a tracheoesophageal fistula and a stenosis of the esophagus. There was deficient peristalsis below the fistula and below the area of stenosis with reflux of contents from the lower esophagus through the fistula and into the trachea, as well as from the stomach into the esophagus and into the trachea. Therefore, this abnormality in motor activity may well be an inherent defect in vagal innervation that is compounded by the surgical procedure. Because of the latter observation (gastro-esophageal reflux) patients with esophageal atresia and tracheoesophageal fistula are placed in a semi-erect position before and after surgery. This position, pre-operatively, may minimize the retrograde flow of gastric juice (which is particularly acid during the first twenty-four hours of life) through the fistula and lessen the degree of pulmonary involvement. After surgical repair, the semi-erect position facilitates the passage of food through the esophagus and into the stomach.

SUMMARY

Cinefluorographic studies of 15 patients operated upon for esophageal atresia, with or without tracheoesophageal fistula, have demonstrated disturbed motor activity of the esophagus. This, when marked, may lead to recurrent aspiration pneumonia and favor the incarceration of foreign bodies in the esophagus. Its observation, pre-operatively, in a patient with tracheoesophageal fistula and esophageal stenosis, raises the question of an inherent vagal deficiency. The semi-erect position is preferred for the patient awaiting surgery, as well as for the postoperative patient.

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ABERRANT INSERTION OF A PULMONARY VEIN INTO THE LEFT ATRIUM SIMULATING AN INTRINSIC LESION OF THE ESOPHAGUS

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AMONG the many congenital malformations of the heart and great vessels, anomalous drainage of the pulmonary veins into the systemic venous circulation is a well-recognized entity. If the anomalous drainage affects the total pulmonary blood flow, life is possible only if there is an associated intracardiac shunt, commonly an atrial septal defect. Partial drainage of one lung or of an individual pulmonary vein into the right auricle may be present as an isolated finding. With the development of cardiac catheterization and angiocardiography, anomalous pulmonary venous drainage has become a relatively common diagnosis.

Other rare abnormalities of the pulmonary veins have been described which primarily concern variations in the total number of veins emptying into the left atrium. This is generally ascribed to the degree of resorption into the atrial wall of the original common pulmonary venous trunk into which the individual pulmonary vessels converge. It is conceivable that variations in the degree of resorption might not only affect the number of pulmonary veins but also their place of insertion into the atrial wall. The result would be an "aberrant" venous insertion.

The authors define aberrant venous return as a condition in which a pulmonary vein inserts into its normal chamber, the left atrium, but the course of the vein varies from the usual pattern. Anomalous venous return represents a deviation from the natural order of embryologic development in which the pulmonary vein inserts outside of the left atrial chamber, compromising the physiologic function.

The following report concerns an aberrant insertion of a left pulmonary vein into the left atrium which, by its position, produced unusual clinical symptoms and roentgen findings.

REPORT OF A CASE

A forty-nine year old white female was first admitted to Lebanon Hospital in October, 1958, with a five month history of precordial pain, substernal in character, radiating to the back and left side. This was associated with episodes of dysphagia, retching, vomiting and, occasionally, syncope.

The past history revealed numerous hospital admissions, including one in January, 1939, for acute epigastric pain associated with tarry stools. Thorough examination at that time failed to demonstrate a causative lesion. In recent years she had been treated for diabetes mellitus, arteriosclerotic heart disease with mild coronary insufficiency and a persistent tachycardia of 120–130 per minute. Further history, after admission, disclosed that the symptoms of dysphagia had been present for many years. She described frequent episodes consisting of a sudden onset of dysphagia, followed by tachycardia, dyspnea, vertigo and incipient syncope. These attacks sometimes occurred during sleep and particularly when she was on her left side. The patient had developed a persistent nonproductive cough a few months prior to the first admission, with one equivocal episode of hemoptysis following a bout of coughing. An intensification of the intermittent dysphagia accompanied these symptoms. Just prior to this admission, the patient was awakened from sleep by severe precordial pressure, dysphagia, retching and collapse, which led to the initial investigation in October, 1958.

Physical examination, on admission, disclosed the blood pressure to be 100/60, with a regular sinus rhythm and a tachycardia of 120

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per minute. A Grade II systolic murmur was heard over the aortic and pulmonic areas with an accentuation of the second pulmonic sound. A mild exophthalmos was present; the thyroid was not palpable. The remainder of the physical examination was within normal limits. Laboratory findings revealed the blood chemistry, blood count and sedimentation rate to be within physiologically normal limits. The electrocardiograms did not point to an acute cardiac episode, but were consistent with arteriosclerotic heart disease and coronary insufficiency. Roentgenograms of the chest were within normal limits. Radioactive iodine uptake studies were normal.

The patient's stay in the hospital was uneventful and she was discharged, at her request, ten days after admission. During the following two months the patient experienced intermittent episodes of dysphagia and retching, which were not severe. She was maintained on digitalis and oral diuretics because of early signs of congestive cardiac failure. At the beginning of December, 1958, the patient developed bouts of right upper quadrant pain, at intervals of two to three days, which increased in severity. These appeared to be unrelated to her previous complaints of dyspnea, dysphagia and the cardiovascular symptomatology, and led to the second admission in December, 1958.

Physical examination, laboratory and electrocardiographic findings were essentially unchanged. The cardiac symptoms were easily controlled with bed rest and medication. Roentgen studies were requested for possible hiatus hernia, peptic ulceration or gallbladder pathology, since the clinical picture suggested a subdiaphragmatic cause for the symptoms.

The initial gastrointestinal series disclosed a large impression upon the anterior wall of the esophagus in the region of the left atrium, with a moderate delay in the passage of the contrast medium at this point. The remainder of the examination revealed the esophagus, stomach and duodenum to be within normal limits. Small bowel examination appeared normal. A repeat examination of the esophageal lesion primarily suggested an intrinsic lesion of the esophagus, intramural in type, due to the overhanging edge on the inferior margin of the lesion (Fig. 2, *A* and *B*). The appearance of the lesion and the clinical evaluation did not suggest simple enlargement of the left auricle. Chest roentgenograms disclosed no abnormality. On esophagoscopy, the esophagoscope passed into the stomach with ease and revealed a normal esophageal mucosa with no evidence of displacement or of external pressure. Laminagraphy of the posterior mediastinal area, with a barium filled esophagus, revealed intimate association of the left auricular appendage with the esophageal defect. The possibility of a localized lesion of the left atrium or appendage was suggested as the probable causative factor.

FIG. 1. Anteroposterior angiocardiogram showing a left pulmonary vein entering the superior aspect of the left atrium somewhat more medially than is usually seen.

Angiocardiography was performed in the frontal and lateral projections using 30 cc. of 90 per cent hypaque injected into the right ventricle through a No. 8 Lehman catheter. The right ventricle appeared minimally dilated. The pulmonary arteries revealed a normal distribution and normal cycle of filling and emptying. Figure 1, taken anteroposteriorly at approximately four seconds after the commencement of injection, shows one of the left pulmonary veins inserting into the superior aspect of the left atrium somewhat more medially than is usually seen. The vein appears dilated and larger in caliber than any of the other visualized pulmonary veins. To study the possibly aberrant course of this vein, angiocardiography was performed in the lateral projection with a barium filled esophagus. Figure 2*A*, taken approximately one second after the commencement of injection, reveals excellent visualization of the right ventricle and right

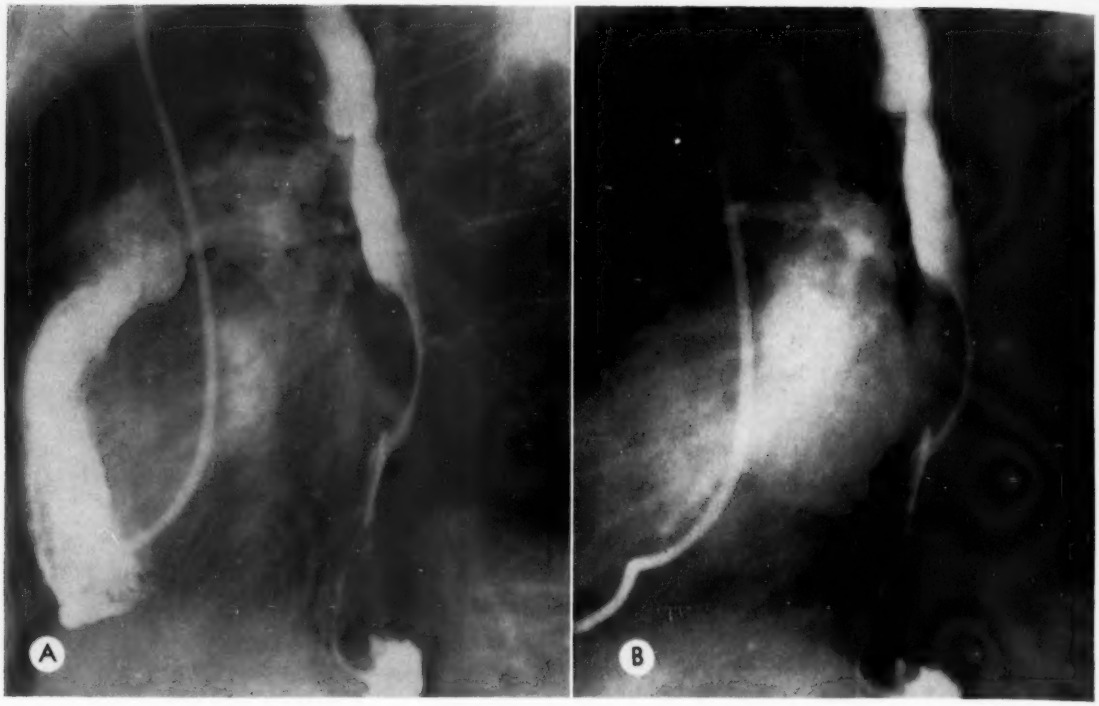

FIG. 2. Lateral angiocardiograms at (*A*) one second and (*B*) four seconds after injection of the contrast medium to show the aberrant course of the left pulmonary vein.

ventricular outflow tract. There is no contrast medium in the area of the esophageal defect. Figure 2*B*, four seconds after the commencement of injection, demonstrates the large left pulmonary vein to be passing over the superior aspect of the left atrium and inserting into the posterior aspect at approximately the midpoint of the chamber. The vein occupies the entire defect in the esophagus and thus can be assumed to be the cause of the defect.

It was felt that this finding satisfactorily explained the symptomatology and the defect noted on the esophageal study. Surgery was not advised to correct this condition. The reason for the attacks of right upper quadrant pain was provided by the results of intravenous cholangiography, which disclosed a stone impacted in the cystic duct. Cholecystectomy relieved these symptoms. Following surgery, fluoroscopic and roentgenographic examinations of the esophagus did not disclose any change in the extent or appearance of the indentation caused by the aberrant insertion of a left pulmonary vein into the left atrium.

The symptoms which initiated the investigation still recur intermittently, but are not incapacitating. Knowledge of the causative factor has relieved the patient's anxiety.

DISCUSSION

Whereas the literature contains numerous reports of anomalous pulmonary venous drainage (complete or partial) into the right atrium or its tributaries, abnormal insertion of a pulmonary vein into the left atrium, with normal drainage, has not been recognized as a clinical entity.

The seemingly unrelated variations that occur in the pulmonary venous drainage have been well understood from the study of the embryology of the pulmonary vascular tree.[2] Similarly, variations in the number of pulmonary veins draining into the left auricle are readily explained if we remember that the pulmonary venous plexus is composed of individual veins which drain the various lobes of the lung buds and which then form a common pulmonary vein entering the left atrium dorsally. Usually, this main pulmonary vein and its branches are incorporated into the dorsal aspect of the left atrium during the formation of this chamber, so that two branches open into the left atrium on either

side. If the common pulmonary vein is incorporated to a lesser degree, single instead of paired vessels may be encountered, and vice versa. Thus Brantigan[1] describes seven varieties of venous insertion. Although occasionally seen at the operating table, this abnormality has, to our knowledge, not been associated with clinical symptoms.

The abnormality visualized in our patient differs from those described by Brantigan as there occurred a cross-over of the left pulmonary vein, which, instead of entering the left atrium on the left, entered it to the right of the midline. As in abnormalities of the aortic arch, such as an arteria lusoria, the importance of this anomaly depends upon whether it produces symptoms. While with an arterial lesion the symptoms are unquestionably those of pressure by the artery on neighboring structures, the symptoms produced in the case of a "vena lusoria" may not only be those of pressure by the vein on neighboring structures but also those of pressure by the neighboring structures on the vein. Due to the low pulmonary venous pressure, the latter condition would appear to be more likely giving rise to symptoms of pulmonary congestion.

The esophageal studies in our patient were performed because of a presenting symptom of dysphagia. The roentgenologic finding on barium swallow was that of an esophageal defect which simulated an intramural, extramucosal lesion of the esophagus. The differential diagnosis was between such an esophageal lesion, bizarre enlargement of the left atrium, tumor of the pericardium and unusual enlargement of the esophageal lymph nodes. The negative esophagoscopy suggested that the defect was produced by extrinsic pressure which could be relieved by pressure of the esophagoscope and this, in turn, suggested a vascular anomaly.

It is surprising that the presenting symptom, dysphagia, was caused by the aberrant course of the pulmonary vein. As stated above, the low pressure pulmonary system would appear more likely to bear the brunt of the pressure with resultant pulmonary congestion. However, the defect seen on barium swallow and which could not be visualized on esophagoscopy was conclusively proved by angiocardiography to be a pulmonary vein crossing in front of the esophagus.

The authors have knowledge of 1 case with similar findings which was operated upon in a New York hospital for a probable leiomyoma of the esophagus. At surgery, an aberrant pulmonary vein causing the esophageal defect was found.

Obviously, this diagnosis can only be made if the opacification of the vein can be seen by angiocardiography. Therefore, this procedure should be considered when the clinician is faced with an unusual defect of the esophagus in the region of the left atrium and a negative esophagoscopy.

SUMMARY

1. Variants in the insertion of pulmonary veins into the left atrium can be easily explained on an embryologic basis.

2. The possibility of such an aberrant insertion causing symptoms of dysphagia is illustrated by the case reported.

3. Since anomalous drainage of pulmonary veins into the systemic venous circulation is known as anomalous pulmonary venous return, the variant described above has been terned "aberrant insertion of the pulmonary vein into the left atrium." It is felt that this is probably a frequent occurrence, although causing symptoms rather rarely.

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LIGNIBEZOAR

REPORT OF A CASE

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BEZOARS have been the source of much interest for many centuries. Indeed, in the 1500's, and possibly even before, an animal bezoar was highly valued for the great powers that it was thought to contain. It was believed that, if the owner were to wear it as a charm or to ingest a particle of it, he would be protected from or cured of a wide variety of maladies, such as epilepsy, plague, headache, dysentery, leprosy, and vertigo. In addition, it would neutralize any poison and would be an effective agent against hemorrhage. In return for one of these wonderful stones, great sums of money changed hands. They became more valuable than gold, silver, and precious jewels.

Man's only interest in bezoars was as a source of miraculous remedy until the late 1700's at which time the first human bezoar was found. Over the years since then, man's superstitious interest in bezoars has given way to an interest in bezoars as a medical curiosity. Though there are about 400 cases reported in the literature, the majority of authors still considers them a rarity.

The definite diagnosis of a bezoar on the basis of history and physical examination alone is unusual and the best that can be hoped for is its inclusion in the differential diagnosis. The patient will rarely volunteer the information that he has eaten hair, persimmons, or other substances related to bezoar formation and it is unusual for the clinician to ask such questions. The symptoms of anorexia, epigastric distress, pain or tenderness, nausea, vomiting, abdominal mass, weakness, weight loss, constipation, and diarrhea, alone or in combination, common to bezoars, are certainly not pathognomonic and could well point to any one of many gastrointestinal disorders. If intestinal obstruction, gastric ulcer (which is usually on the lesser curvature), or perforation of an ulcer should occur as a result of the bezoar, the patient will present with the usual symptoms of that particular gastrointestinal complication.

Laboratory studies may help to point the way to a correct diagnosis. The majority of cases are accompanied by a mild leukocytosis and a slight secondary anemia. In the case of a trichobezoar, gastric analysis frequently reveals low acidity, while normal or high acidity is usually found in cases of phytobezoars. In addition, bits of the component material may be obtained with the specimens for gastric analysis. Also, stool examination may reveal particles of the offending substance.

The roentgenographic examination will usually reveal the correct diagnosis. It can be said that the characteristic roentgen findings, by both roentgenography and fluoroscopy, are pathognomonic. At the fluoroscopic examination, the ingested barium meal will be seen to outline the stomach walls leaving a radiolucent filling defect within the lumen of the stomach. At the same time, it may be noted that the stomach is distended. As more of the barium meal passes into the stomach, the general outline of the filling defect remains but it will begin to take on a striated or mottled appearance as the barium fills the small spaces between the particles of matter making up the bezoar. The general outline of the filling defect will usually match that of the stomach itself. The mass is frequently movable on palpation and, when the stomach is filled with barium, it may be pushed toward the gastric fundus so that a portion of this striated or mottled mass will protrude into the gastric air bubble above the air-fluid level. The roentgenograms will confirm the fluoroscopic findings. It is frequently helpful to obtain a roentgenogram of the abdomen several hours after the

initial examination, when the stomach has emptied the barium contents, since it will frequently reveal in the region of the stomach a mass which has a striated or mottled appearance due to barium temporarily trapped in crevices between the consituent particles of the bezoar.

Many fine articles concerning bezoars can be found in the literature. Rather than to attempt to repeat what has been so adequately written by others, the foregoing was designed as a brief summary of bezoars in general. For greater detail, the reader is referred to the reports of Thorington,[2] DeBakey and Ochsner,[1] Tondreau and Kirklin,[3] and others who have contributed to our knowledge of bezoars through the medical literature. The primary purpose of this report is to present a case history of an unusual type of bezoar, one consisting of wooden splinters.

REPORT OF A CASE

The patient was a well developed, well nourished eighteen month old white male child. The parents noted him chewing on parts of the crib railing. Eventually, about ½ inch of the wood had been removed most of the way around the crib with no evidence of the pieces to be found. Two or three days after this wood chewing was first noticed, the patient began to pass wooden splinters in his stools. At this point, the crib railing was covered with tape to prevent further chewing. He continued to pass 3 to 5 loose stools per day, all with considerable mucus and bits of wood. On one occasion he vomited pieces of wood up to 2½ inches long. During ths time, he had continued to eat well. The parents stated that the child had no apparent abdominal pain but he did cry when moving his bowels. The patient was seen by the family physician who suggested that he be admitted to the hospital to determine the amount of wood remaining in his gastrointestinal tract.

The child was admitted to the Robert Packer Hospital in Sayre, Pennsylvania, on March 25, 1960, approximately three weeks after he was first noticed chewing on his crib. He continued to pass splinters in his stools (Fig. 1) but had no more episodes of vomiting after the original single occasion.

The system review was negative and it was

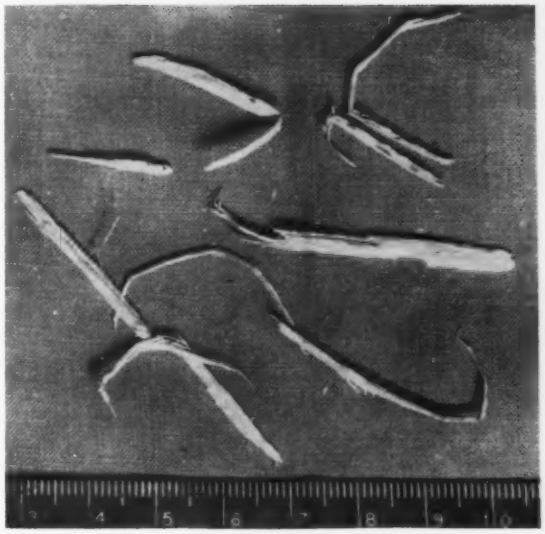

FIG. 1. A few of the wooden splinters found in the patient's stool. The scale is in centimeters.

determined that he had been gaining weight, although slowly. He had been hospitalized at Robert Packer Hospital in November, 1959 for bronchopneumonia which had responded well to treatment. There were no known allergies. He was eating table foods and taking vitamins.

The physical examination revealed a normal, alert, cooperative child weighing 31 pounds. No abnormalities were found.

A blood count obtained on the day of admission reported 8.9 gm. of hemoglobin, hematocrit 31 per cent and a white blood cell count of 6,650 with a differential of 44 per cent neutrophils and 56 per cent lymphocytes.

Also on the day of admission, a gastroduodenal examination was requested of the Radiology Department. At fluoroscopy, after ingestion of a barium mixture, a rather large filling defect was noted within the lumen of the stomach; it was roughly triangular in shape with the apex projecting into the gastric antrum. The mass was freely movable and the fluoroscopist gained the impression that it floated on the ingested barium-water mixture. The walls of the stomach appeared normal and barium was seen to pass freely from the stomach into the duodenum. The duodenal bulb appeared to be within normal limits. The spot roentgenograms revealed a negative filling defect within the gastric lumen (Fig. 2). This defect appeared to contain linear striations of increased density apparently due to the ingested barium. A roentgenogram obtained one

FIG. 2. Spot roentgenogram, obtained during the fluoroscopic examination, showing the stomach filled with barium and the bezoar represented by a mottled, striated negative filling defect.

hour after the initial examination revealed the barium meal passing freely through a normal small intestine. Another roentgenogram taken four hours after the barium had been ingested

FIG. 3. Roentgenogram obtained four hours after ingestion of the barium meal showing the barium impregnated bezoar in the stomach and the barium meal in the large and small intestine.

FIG. 4. The bezoar, consisting of wooden splinters, which was removed from the patient's stomach at surgery. The scale is in centimeters.

revealed a barium coated foreign body within the stomach, with no residual free barium (Fig. 3). The roentgen diagnosis was reported as a foreign body within the stomach having the appearance of a bezoar.

A single scout roentgenogram of the abdomen was obtained three days after the initial examination; it once again showed the barium impregnated mass in the region of the stomach. It appeared to be of the same size as originally noted. A period of forty-eight hours passed during which no splinters of wood were found in the stool so it was decided that the bezoar should be removed surgically.

On March 29, a gastrotomy was performed. At operation, a large mass was felt in the stomach and, after opening the stomach, a large ovoid concretion composed of numerous wooden splinters cemented together by thick mucoid material was removed (Fig. 4). The mass measured 9.5 cm. in length by 3.5 cm. in width. The wooden splinters averaged 6 to 9 cm. in length. A thorough search was made for other splinters, but none were found, and the stomach was closed.

The patient did well postoperatively and no complications were encountered. He was discharged on the ninth postoperative day, April 7, 1960.

DISCUSSION

Bezoars have been divided into four categories on the basis of their constituent materials. Trichobezoars are those composed of hair. They range from human hair through animal hair to string and thread. Phytobezoars are composed of various food materials of plant origin. They may consist

of stems, fibers, leaves, roots, and seeds. The most common in this category are persimmon bezoars. Trichophytobezoars are those composed of a combination of the materials in the above two categories. The fourth category includes miscellaneous bezoars and concretions which have been reported as consisting of asphalt, shellac, sand, and other substances.

The bezoar described in this presentation would most likely be classified in the fourth category as a miscellaneous bezoar. True, it is of plant origin but it certainly is not a food substance and, therefore, not a true phytobezoar. It is suggested that bezoars of this nature might be termed lignibezoars.

SUMMARY

A brief summary has been presented of the history, symptoms, and laboratory and roentgen findings of bezoars. A case history has been reported of an unusual type of bezoar consisting of wooden splinters with the suggestion that it might be termed a lignibezoar.

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PRIMARY RHABDOMYOSARCOMA OF THE STOMACH AND ESOPHAGUS*

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PRIMARY rhabdomyosarcoma of the upper gastrointestinal tract is a rare tumor. Stout and Lattes[6] reviewed 4 cases of rhabdomyosarcoma primary in the esophagus, and Stout[7] has seen 2 cases of rhabdomyosarcoma involving the stomach. Geschickter,[2] Gordon-Taylor,[3] and Pack and Ariel[5] in their extensive reviews of striated muscle tumors do not include any cases in which the esophagus or stomach was the organ of primary involvement.

The roentgenologic, radiotherapeutic, surgical and pathologic findings in 2 cases of rhabdomyosarcoma that involved the stomach and upper esophagus respectively form the basis of this report.

Rhabdomyosarcomas occur in both sexes equally and at any age. Trauma does not appear to be a causative factor. Although the exact etiology is unknown, the congenital theory appears to be most tenable. Geschickter[2] has pointed out that within the first month of embryonic life the primitive digestive tube is partially invested with a sheet of mesoderm from which a part of the body's skeletal musculature is derived. Also, mature striated muscle cells are completely differentiated end products and are unable to undergo cell division. The growth characteristics of these tumors are extremely variable.[3]

Rhabdomyosarcomas tend to metastasize quite early to the lungs via the blood stream. They may, less commonly, disseminate by way of the lymphatic system. Pack and Ariel[5] who show a 33 per cent five-year cure rate in their series, feel that surgery is the treatment of choice; however, the judicious use of radiation therapy has a definite place in the over-all management of the patient with this lesion.

REPORT OF CASES

CASE 1. A three year old Negro male was seen at Henry Ford Hospital with a six month history of painless swelling in the right neck. Physical examination was completely negative except for a 4×6 cm. freely movable, spherical, cystic mass located in the anterior triangle of the neck.

Surgical exploration of the neck on June 13, 1960 disclosed a tumor which did not appear to arise from, nor have any connection with, the surrounding musculature. It was completely excised, and the pathologic interpretation of the frozen sections was "cervical lymph node, almost completely replaced by tumor."

Subsequent paraffin sections of the tumor tissues stained with hematoxylin and eosin showed a highly cellular tumor with a scanty fibrous tissue stroma. There were clusters, sheets and clumps of both, small round cells with minimal cytoplasm and larger spindle shaped cells, some of which were in an alveolar pattern. These latter cells showed polar elongation of the cytoplasm into homogeneous pink processes in which no fibrils or striations could be definitely identified. Imprint preparations showed fairly uniformly distributed chromatin particles in the nuclei. Of special interest was the identification of giant, intensely pink, glassy appearing nucleoli. The final diagnosis was rhabdomyosarcoma partially replacing a lymph node.

Numerous physical, laboratory, and roentgen examinations did not demonstrate the primary tumor site. The child was discharged on June 19, 1960, to be followed in the out-patient clinic.

One month later he was re-admitted with a two week history of anorexia, intermittent vomiting of yellow bile-like material and a four pound weight loss. There was no history of hematemesis or melena.

Physical examination at this time revealed a firm, nontender, well circumscribed abdominal

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mass projecting from beneath the left costal margin. Bowel sounds were normal, and the abdomen was not distended. The chest was clear. Laboratory studies were noncontributory. The child developed symptoms of increasing high intestinal obstruction. Roentgen studies of the abdomen (Fig. 1), urinary system (Fig. 2), and upper gastrointestinal tract (Fig. 3) were obtained.

Because of the roentgen demonstration of a large gastric tumor, a laparotomy was performed on July 27, 1960. A huge mass was immediately encountered arising from and involving the upper two-thirds of the stomach. The gastric mass was contiguous with large indurated lymph nodes located in the pyloric, celiac and porta hepatis areas. The pylorus itself was infiltrated and partially obstructed. Since curative surgery was not thought possible, a gastrojejunostomy was done and multiple biopsies were taken.

The patient's immediate postoperative course was satisfactory. Because of the large mass encroaching on the lumen of the proximal stomach, further obstruction was anticipated. Radiation therapy was therefore initiated in an effort to reduce the tumor mass. A minimum tumor dose of 4,500 r was planned, and, if at the end of this time the patient was tolerating the radiation satisfactorily, the tumor dose would be carried to 6,000 r. Cobalt 60 was used with a daily tumor increment of 200 r. Unfortunately, during the next two weeks the

Fig. 2. Case 1. An excretory urogram shows normal renal outlines, position and collecting systems. The mass in the left upper quadrant is sharply outlined.

Fig. 1. Case 1. Supine roentgenogram of abdomen demonstrating a large homogeneous soft tissue density in the left upper abdomen and deformity of the gastric air bubble.

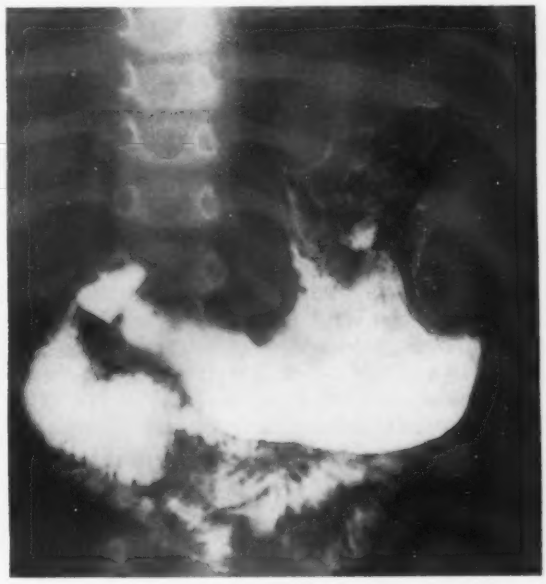

Fig. 3. Case 1. A roentgenogram of the stomach demonstrates a large gastric mass producing a filling defect in the fundus and body of the stomach. There is generalized mucosal destruction.

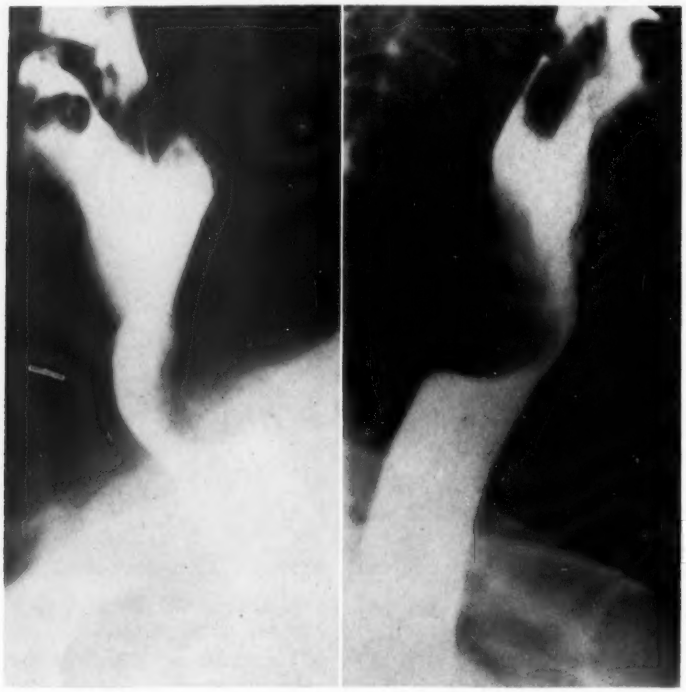

FIG. 4. Case 11. Esophagrams show a smooth, intramural defect in the posterior cervical esophagus.

patient began to accumulate ascites and developed urinary retention. Therapy had to be discontinued on August 18, 1960 after a tumor dose of 2,200 r had been delivered. The child expired ten days later.

Significant autopsy findings were limited to the abdomen. The entire stomach was involved by a huge tumor mass which extended to the second portion of the duodenum, the left lobe of the liver, and the undersurface of the left hemidiaphragm. In addition, there was direct extension of tumor into the pancreas. Metastatic disease was not found. Microscopic examination of the tumor tissues revealed a pattern entirely similar to that seen in the cervical lymph node. There was no histologic evidence of any nuclear or cellular change that could be attributed to radiation.

Comment. Malignant tumors of the stomach are very unusual in young children.[1] McNeer[4] in reviewing the world literature found 19 cases of gastric cancer in the age period from birth to fifteen years. The sites of malignancy in children differ from those in adults, and, although no part of the body is exempt, 80 per cent of all malignant tumors in young children occur in the adrenals, kidneys, eyes, blood, brain or bones.[8] There is little correlation between the histologic characteristics of a particular tumor and its biologic behavior.

There was a time lapse of six weeks from the initial physical examination and cervical lymph node excision to the day of abdominal exploration. In this time the large epigastric mass became apparent. Such rapid enlargement would be typical of the "hurricane" growth described by Gordon-Taylor.[3] The factor, or factors stimulating such sudden and rapid tumor growth remain unknown.

The first and only known distant metastasis in this case was to a cervical lymph node. There was also extensive spread of the tumor to the celiac and portal lymph node structures at the time of laparotomy and autopsy. The more common blood-borne metastases were not found.

It is not surprising that the tumor did not show any clinical regression or subsequent pathologic changes attributable to the radiation therapy. Unfortunately, the patient's poor condition made radiation

therapy impossible after a tumor dose of 2,200 r was given. It might be argued that the patient did not live long enough for any radiation induced tumor changes to become manifest. The general impression gained from reading the scant literature on this subject is that these tumors are almost always radio-incurable. However, they are frequently radio-responsive to a moderate dose of therapy.

CASE II. A fifty-five year old white male entered Henry Ford Hospital on August 22, 1960 with a two and one-half month history of painful, difficult swallowing. He denied nausea, vomiting, hematemesis, significant voice change, or melena. He had a twenty pound weight loss in three months.

Physical and laboratory findings were noncontributory. Examination of the ears, nose, and throat, including direct laryngoscopy, was negative as was thyroid scanning. Roentgenograms of the esophagus (Fig. 4) were obtained and because of the findings, esophagoscopy and biopsy were performed. The esophagoscopist described mild indentation of the posterior cervical esophagus but the biopsy did not reveal any abnormality.

Surgery subsequently showed the defect to be a hard, 4 by 4 cm. encapsulated mass lying within the musculature of the posterior esophagus just below the level of the cricopharyngeal muscle. The tumor was completely removed.

Pathologic examination of the surgical specimen revealed a bizarre, compact pattern of giant cells with peripheral nuclei, elongated strap cells, small round cells, and "racquet" cells intermixed with striated muscle fibers. There was a scanty amount of eosinophilic cytoplasm. PAS with and without diastase digestion showed the presence of a large amount of glycogen in the tumor. These latter findings and the absence of wiry reticulum between the tumor cells indicated the tumor to be of striated muscle origin. The final diagnosis was rhabdomyosarcoma.

Comment. The embryogenesis of the tumor in this case is very likely the same as in Case 1. The growth characteristic is probably one of slow growth with sudden acceleration. Again, the growth stimulus remains unknown. Because of complete surgical enucleation of the tumor, radiation therapy was not indicated at this time. The patient has remained symptom free.

SUMMARY

Two cases of primary upper gastrointestinal tract rhabdomyosarcomas have been presented. Our experince in regard to the roentgen findings, treatment and pathology of this very rare tumor is discussed.

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The authors wish to thank Dr. Roy Patton for the interpretation of the pathologic material. The helpful suggestions of Drs. William R. Eyler and Robert J. Bloor are also gratefully acknowledged.

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GASTRODUODENAL INTUSSUSCEPTION

By SVERRE GRÖNLIE *and* BJARNE ROSSELAND
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INTUSSUSCEPTION in the intestinal tract occurs rather frequently, and gastroduodenal intussusception is not rare, although the diagnosis is seldom made preoperatively. The first report of roentgenologically demonstrated gastroduodenal invagination was that of Lönnerblad[1] in 1933, who presented 2 cases.

In that report he described in detail the roentgenologic criteria for the diagnosis of intussusception, *i.e.*, the characteristic contour of the mucosa and the distinctive appearance of the apex of the intussusceptum when contrast material is employed. The same criteria are valid for the relatively common ileocolic intussusceptions seen in children. In children these invaginations are rarely caused by pathologic alterations, while in adults they are almost always the result of pathologic processes, such as polyps, tumors, foreign bodies, etc.

In 1939 Zdansky[4] reported 1 case, and in 1955 Mauthe and Zwicky[3] reported 2 cases of gastroduodenal invagination which had been diagnosed roentgenologically. Marshak *et al.*[2] described in 1951 a case which was intrepreted roentgenologically as a tumor in the descending part of the duodenum, but which proved on operation to be an intussusception of a tumor from the greater curvature in the pyloric antrum. In their paper, the authors gave a summary of a number of cases of gastrointestinal invagination, but they found only 2 cases in which the diagnosis had been made preoperatively, and these were the 2 cases of Lönnerblad.[1]

It is probable that the diagnosis is rarely made by the roentgenologist because, in most instances, the invagination is a labile condition in which the intussusceptum moves in and out. There appears to be a certain periodicity in the symptomatology which supports this assumption. The typical findings are most readily obtained when the intussusceptum is incarcerated, with the pylorus as the hernial orifice, or when the patient is examined by chance just at the moment when the intussusception is present.

The present report is of a case in which the intussusceptum was incarcerated.

REPORT OF A CASE

A seventy-eight year old man, pensioned, was transferred to Sentralsykehuset for Östfold i Fredrikstad, Medical Department, from another hospital where he had spent a little less than three weeks. For the preceding two years he had suffered from a dyspepsia, but not to such an extent that he had sought medical advice until he suddenly had hematemesis about seven weeks before admission to Sentralsykehuset. He had subsequently twice had hematemesis which was relatively severe and was followed by melena. He grew debile, consulted a physician, and was admitted to the nearest hospital. Here the initial treatment was blood transfusions and his general condition gradually improved. Rectoscopy and roentgen examination of the colon and stomach revealed no definite pathologic signs except for sigmoid diverticula. However, as his condition later grew worse, he was transferred to the Medical Department at Sentralsykehuset.

On admission the patient was pale, slightly cyanotic, and complained of pains around the anal region and downward in the left thigh. Clinical findings included: pulse rate, 80 and regular; blood pressure, 135/75; hemoglobin, 7.59 gm. per 100 ml. of blood; heart, uncharacteristic systolic murmur; electrocardiogram, normal; abdomen, normal intestinal auscultation, soft, not sensitive, no intestinal tautness, and no palpable tumor.

Roentgenograms from the previous hospital admission were available and careful scrutiny revealed in one of them an alteration in the descending part of the duodenum, which indicated the need for further examination. On the new roentgenograms the typical contours seen in intussusception were found as well as a pronounced apex of the intussusceptum (Fig.

Fig. 1. Pronounced longitudinal stripes—the stalk—surrounded by rings of contrast material in two layers.

1 and 2). On the basis of these findings the Roentgen Department made the diagnosis: gastroduodenal intussusception. The patient was transferred to the Surgical Department and was operated at once.

The roentgenologic diagnosis was confirmed. We cite from the surgeon's (Rosseland) description: At first inspection, both the stomach and the duodenum appeared normal except that the pylorus seemed wide. Palpation relatively far down on the descending part of the duodenum revealed a fairly soft tumor, almost the size of a tennis ball. Extending from the tumor upward through the pylorus and to the posterior wall of the stomach a rope-like structure was palpable. There was a deep indentation in the fundus, the bottom of which could not be reached with a finger. It thus appeared

Fig. 2. Shortening and obliteration of the lesser curvature with indentation in the fundus (arrow, upper right) and an egg-shaped tumor in the descending part of the duodenum with pronounced apex invagination (arrows, left).

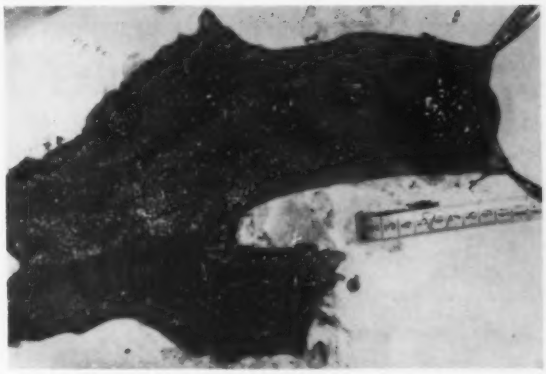

Fig. 3. The gastric specimen spread open with the tumor attached to the upper part of the anterior wall of the fundus.

that the tumor had its origin in the upper part of the fundus. Attempts to push the tumor into the stomach manually were of no avail, so gastrotomy was performed. A mucous stalk was revealed, and by pulling on this it was possible to draw the tumor up. There was a necrotic, bleeding area on the upper surface of the tumor, which had a broad base arising from the anterior wall of the stomach. The bleeding was rather profuse. As the tumor was located so high in the stomach, a subtotal gastrectomy was performed with the usual anastomosis. The portion of the stomach which was removed (Fig. 3 and 4) measured 30 cm. along the side of the greater curvature, and 15 cm. on the side of the lesser curvature. The specimen weighed 340 gm.

Histologic diagnosis: leiomyoma with necrosis.

The postoperative course was uncomplicated.

Fig. 4. The same specimen as in Figure 3 with the tumor intussuscepted.

Comment. It is not difficult to make the diagnosis when the characteristic contours are present (Fig. 1 and 2). The shortening of the stomach, the obliteration of the lesser curvature, the widening of the descending part of the duodenum and the marked apex of the intussusception are visible in Figure 2. There is also a central tunnel-shaped defect in the contrast material with axial stripes surrounded by rings (Fig. 1). These longitudinal stripes represent the innermost lumen of the tube—a stalk—while the circular rings of contrast indicate the middle and outer tubes formed partly by swelling of Kerkring's folds and partly by folding of the outer intestinal layer. This description is, on the whole, similar to that presented by Lönnerblad.[1]

SUMMARY

A case of gastroduodenal intussusception diagnosed preoperatively is reported. A short review of 5 cases found in the available literature is given.

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INTUSSUSCEPTION OF A MALIGNANT DUODENAL POLYP*

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THE incidence of polyps of the small bowel is relatively small, but of these about 45 per cent occur in the duodenum. Those reported have been benign. Infrequently, such polyps are complicated by intussusception. Recently, we examined a patient with a duodenojejunal intussusception secondary to a malignant polyp.

REPORT OF A CASE

A fifty-eight year old Caucasian male was admitted December, 1959, with a history of indigestion and occasional bloating of five to six years' duration. For the preceding two months, he had noted episodic distention associated with constipation and rectal bleeding. There was no weight loss. A large polypoid defect had been demonstrated in the duodenum on 2 different roentgen examinations. In view of the more recent symptoms, he was admitted for elective surgery.

His past medical history included a pituitary adenoma in 1955 treated with radiation therapy. Later he developed blindness of the left eye and adrenal steroid deficiency. He was well maintained on hydrocortisone.

Physical findings were within the limits of normal, except for the blindness. Routine laboratory studies were normal.

Roentgen Studies. The first examination was done at another hospital in 1954 at the onset of the patient's abdominal complaints. The study showed a large diverticulum arising from the descending loop of the duodenum, and an oval filling defect in the third portion of the duodenum, measuring 6 by 4 cm., without obstruction (Fig. 1). He was examined again in November, 1959, and the same abnormalities were demonstrated (Fig. 2). The filling defect appeared unchanged since the previous examination five and one-half years earlier. The roentgen interpretation was a benign tumor. Study at our hospital one month later again demonstrated the diverticulum and the tumor, with the additional complication of extensive intussusception (Fig. 3, *A* and *B*).

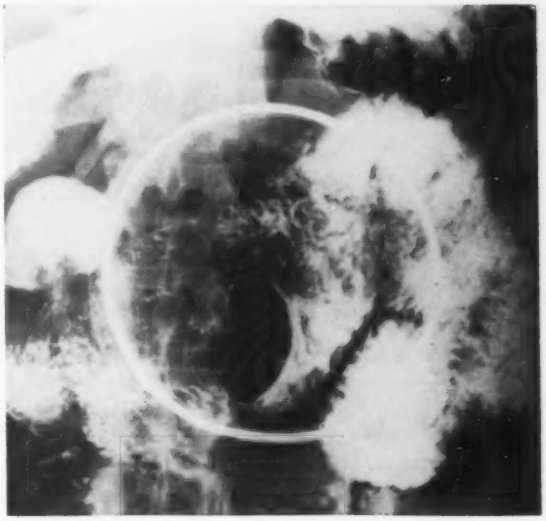

FIG. 1. *May, 1954:* initial study. Note the large diverticulum arising from the descending duodenum, and an oval filling defect in the third portion without intussusception.

The preoperative diagnosis was duodenal adenoma. At surgery, a polypoid tumor approximately 5 cm. in diameter was found in the second portion of the duodenum. Since the muscular coat seemed to be invaded, a frozen section was obtained. Well-differentiated adenocarcinoma was reported. Therefore, a subtotal gastrectomy, partial duodenectomy and antecolic gastrojejunostomy were performed. No other tumor was seen. Since surgery, the patient has done well. When last seen five months after the operation, he was asymptomatic and was working full time.

Histologically, the tumor proved to be a pedunculated adenomatous polyp with a focal area of well-differentiated adenocarcinoma showing limited invasion of the stroma (Fig. 4).

DISCUSSION

Polyps of the small bowel are most fre-

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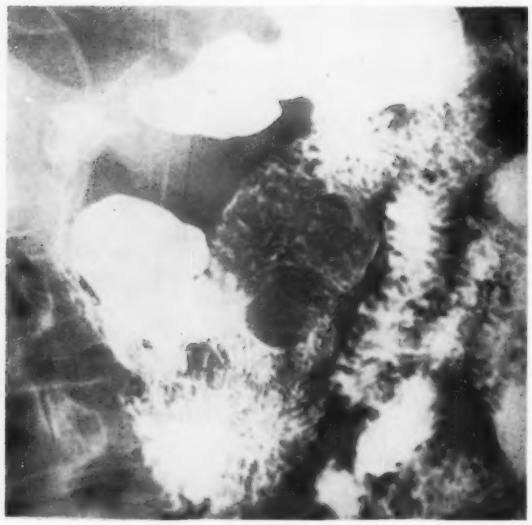

FIG. 2. *November, 1959:* repeat study five and one-half years after Figure 1. The same abnormalities are demonstrated. The filling defect appears unchanged. Once again there is no evidence of intussusception.

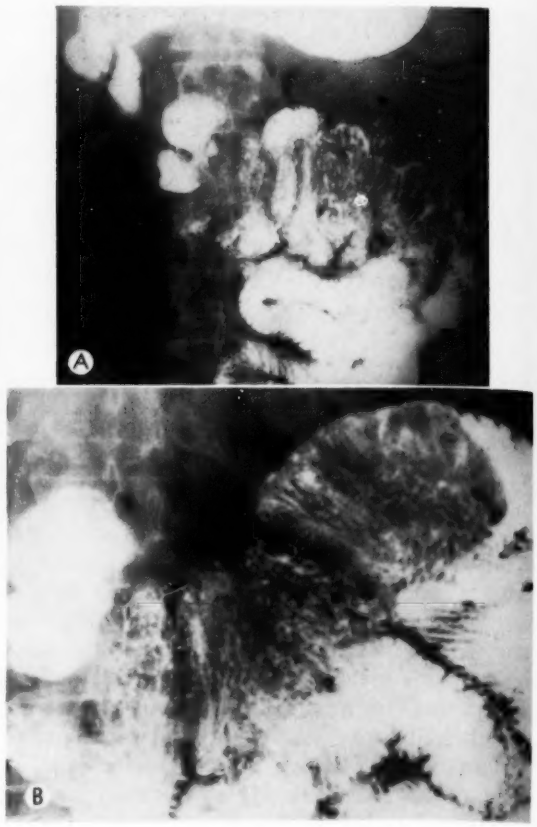

FIG. 3. *December, 1959:* one month after Figure 2. (*A*) The diverticulum and the tumor are again demonstrated, with the additional complication of extensive intussusception. (*B*) Another view of the intussusception enlarged for detail.

quently asymptomatic, and discovered at autopsy. While vague gastrointestinal symptoms occur occasionally, polyps which become symptomatic generally present with obstruction, especially intussusception, or bleeding. River *et al.*,[6] in their comprehensive review of benign small bowel neoplasms, found them to be the most common cause of intussusception in adults. However, this complication is usually of ileal origin. Prolapse of gastric mucosa or polyps with gastroduodenal intussusception are also seen with some regularity, but intussusception of a duodenal polyp is rare. River *et al.* found 13 references to such in the world literature, and since their review, we have learned of 4 others,[2–4] all benign. At least 1 of these, in addition to our case, intussuscepted beyond the ligament of Treitz.

The rarity of duodenal intussusception seems best explained on an anatomic basis. The duodenum differs from the rest of the small bowel in that it has no mesentery. The first portion is somewhat mobile, but the remaining three portions are fixed by the overlying peritoneum to the posterior abdominal wall, liver and pancreas. In addition, the duodenojejunal flexure is held firmly in place by a fibrous and muscular band, the ligament of Treitz.

Roentgen examination is the most valuable method in the diagnosis of benign duodenal tumors, and their differentiation from biliary, pancreatic or peptic ulcer disease. Typically, benign tumors present as well-defined regular filling defects—usually circular, sometimes multilocular. A pedicle is considered by many to favor benignancy. Ulceration may be present, but the mucosal pattern is maintained.

Histologically, the majority of duodenal polyps are papillary adenomas. Of the polyps which intussuscepted, only four exceptions were found—1 fibroadenoma,[6] 2 fibromyomas[6] and 1 myoma.[4]

Malignant lesions of the duodenum are

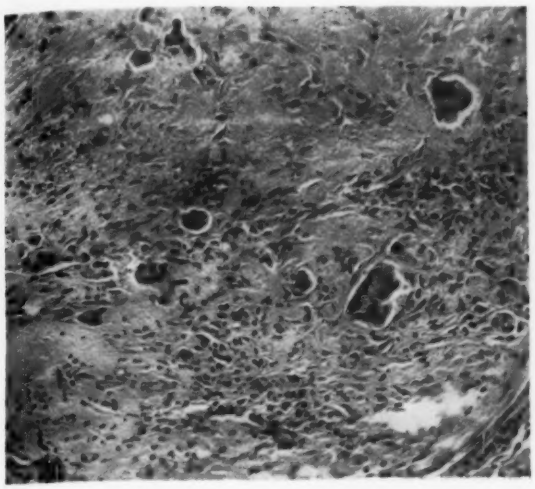

Fig. 4. Section of malignant portion of the polyp showing the presence of malignant cells in vascular channels (×200).

infrequent with an estimated incidence of 0.035 per cent based on a total of 487,695 autopsies.[5] Of these, 60 per cent occur in the periampullary region. The clinical picture correlates with the morphologic characteristics of the lesions. These may be categorized as follows: (1) obstructing lesions, (2) ulcerating lesions, (3) lesions penetrating into the pancreas and (4) lesions encroaching upon the ampulla of Vater. Accordingly, the major symptoms will be (a) vomiting, (b) bleeding, (c) gnawing pain unrelieved by food or alkali and (d) obstructive jaundice. Weight loss often provides the strongest clinical suggestion that the underlying lesion is malignant.

Roentgen examination again is the most valuable procedure in the diagnosis of malignant duodenal tumors. Brenner and Brown[1] outlined the important abnormalities seen in malignant lesions of the duodenum as follows: (1) annular, infiltrating or ulcerative defects, (2) effacement and thickening of the mucosa, (3) irregular polypoid defects which narrow the lumen, (4) dilatation of the duodenum above a constricted area and dilatation of the stomach with gastric retention, and (5) rigidity of the duodenal wall.

The majority of malignant duodenal lesions are adenocarcinomas. The next most common are lymphosarcomas with a ratio for the two of approximately ten to one. Leiomyosarcomas, carcinoids and other tumors are observed very rarely. An estimated 45 per cent of small bowel carcinomas occur in the duodenum, in contradistinction to sarcomas and carcinoids which occur more frequently in other parts of the small bowel. Where histologic detail has been reported, duodenal carcinomas have been papillomatous in configuration. No report of a carcinoma associated with a duodenal polyp was found. The question of malignant degeneration in polyps comes forcibly to mind, but is beyond the scope of this case report.

SUMMARY

1. A case of a fifty-eight year old Caucasian male having a duodenal polyp which intussuscepted and which proved to contain focal adenocarcinoma is presented.

2. Duodenal intussusception is briefly discussed.

3. Benign and malignant lesions of the duodenum are compared in regard to incidence, clinical behavior, roentgen appearance and histology.

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A ROENTGENOLOGIC REMISSION IN ULCERATIVE COLITIS

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ULCERATIVE colitis is generally considered to be a chronic disease characterized by remissions and exacerbations and pursuing a mild course in most, and a rapidly progressive, occasionally fatal, course in others. Complete clinical remissions are not unusual, but it is generally not appreciated that chronic ulcerative colitis may have a complete roentgenologic remission. Generally, the changes of far advanced ulcerative colitis have been regarded as permanent. Indeed, when shortening of the colon, narrowing of the lumen, and stricture formation have occurred, it is difficult to understand how the process could be reversible. The following report illustrates a far advanced case of ulcerative colitis that had a complete clinical and an apparent roentgenologic remission.

REPORT OF A CASE

The patient was a nine year old white female referred to Jefferson Medical College Hospital on January 30, 1948, with a diagnosis of ulcerative colitis.

Early in 1946, she developed what her parents described as "intestinal grippe," which consisted of fourteen to twenty watery stools per day containing mucus and blood. The episode lasted one week and subsided spontaneously. There were two more occurrences of diarrhea, which subsided without treatment, during the remainder of 1946. In August, 1947, frequent loose bowel movements recurred and gradually increased in severity. Because of the persistence of diarrhea, she was admitted to another hospital in December, 1947, and a diagnosis of ulcerative colitis was made. When she failed to respond to therapy, she was referred to Jefferson Medical College Hospital.

Family history revealed that she was the oldest of four sisters. She was described as above average in intelligence by her mother, was apparently well adjusted, and came from a reasonably happy home.

Physical examination disclosed a thin well-developed white female child who was 54 inches tall and weighed 54 pounds. The temperature was 99.2° F. orally. The remainder of the physical examination was not remarkable. Proctosigmoidoscopy revealed a granular, engorged and extremely friable rectal mucosa, and bleeding occurred with slight trauma. The findings were felt to be characteristic of chronic ulcerative colitis. The laboratory studies were within normal limits. Barium enema examination (Fig. 1*A*) was performed and was interpreted as being consistent with ulcerative colitis. After a slight clinical improvement, the patient was discharged on February 6, 1948.

In April, 1950, the patient was readmitted because of frequent bloody bowel movements. Since her previous discharge in February, 1948, she had had four to six loose stools per day. About six weeks before admission, she had begun to have six to fifteen watery, frequently bloody, stools per day. The physical examination was not remarkable. Proctosigmoidoscopy revealed the perineal skin to be reddened and tender, and the mucosa was thickened, friable, and showed ulceration. Barium enema examination (Fig. 1*B*) showed progression of the ulcerative colitis. The patient was given a course of ACTH, followed by cortisone, and marked improvement was noted with the stools becoming formed and less frequent. After June, 1950, there was steady clinical improvement. Two brief exacerbations of diarrhea in July, 1951, and November, 1952, subsided spontaneously. Frequent proctoscopic examinations revealed a gradual but steady decline in the mucosal inflammation. In November, 1952, the colon showed little change roentgenologically (Fig. 1*C*), but one year later (Fig. 1*D*)

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Fig. 1. (*A*) *January, 1948:* The colon is narrowed and somewhat shortened, and there is complete loss of haustral markings. The edge of the barium column is irregular. (*B*) *April, 1950:* Further shortening has occurred and a stricture is evident at the hepatic flexure. The ileocecal valve is patent, and the terminal ileum also appears to be involved. (*C*) *November, 1952:* Shortening of the colon is still evident but there is more distensibility. Stricture of the hepatic flexure is still present. (*D*) *October, 1953:* The colon has lengthened somewhat, and is more distensible. The stricture at the hepatic flexure is still present.

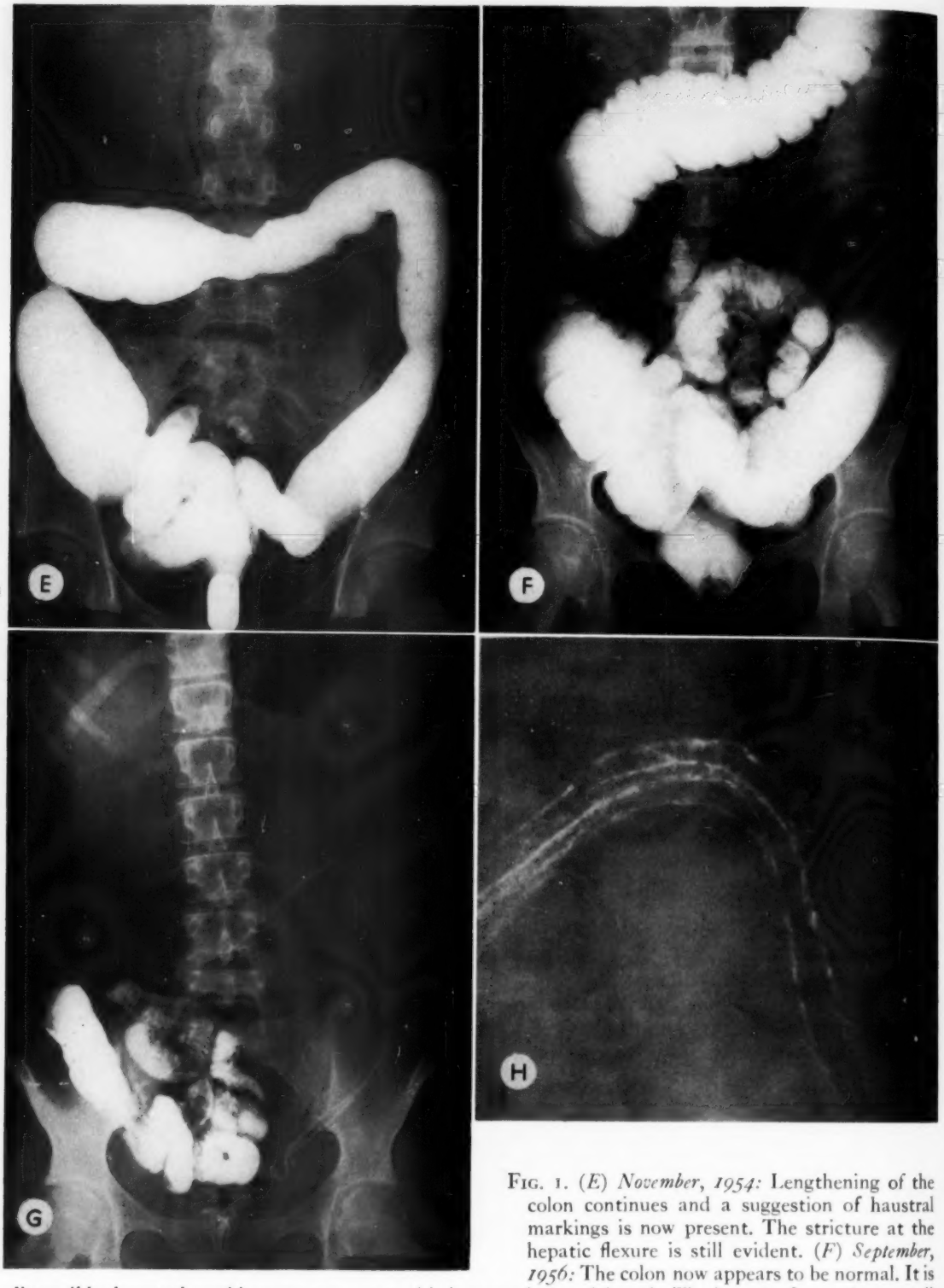

FIG. 1. (*E*) *November, 1954:* Lengthening of the colon continues and a suggestion of haustral markings is now present. The stricture at the hepatic flexure is still evident. (*F*) *September, 1956:* The colon now appears to be normal. It is distensible, haustral markings are present, and it is now of normal length. The hepatic flexure is not well demonstrated. (*G* and *H*) Postevacuation roentgenogram and close-up study of the mucosal pattern at the splenic flexure show a cobble-stone appearance of the mucosa with longitudinal furrowing. The mucosa is abnormal.

the bowel exhibited definite improvement, which continued in 1954 (Fig. 1*E*). She was completely asymptomatic in September, 1956, and no evidence of activity was noted at proctoscopy. The roentgenologic appearance of the colon (Fig. 1*F*) was normal, but in the postevacuation roentgenograms (Fig. 1, *G* and *H*) the mucosal pattern was still definitely abnormal. Since September, 1956, her family physician has reported that she has married, has had an uneventful pregnancy, and has had no exacerbations of ulcerative colitis.

DISCUSSION

The characteristic roentgenologic changes of ulcerative colitis are well known. In the early stages, the alterations are limited to the mucosa. The edges of the barium column appear serrated, and there is hypertonicity. Mucosal ulcerations can be demonstrated in many cases. Later, loss of haustral markings occurs, and eventually the colon shortens and becomes narrow. Stricture formation is not uncommon. Ultimately, the picture is one of a markedly shortened, narrowed, pipestem colon with an irregular mucosal pattern that shows ulceration and pseudopolypoid changes.

In our case, the early roentgen studies (Fig. 1, *A*, *B* and *C*) showed the typical roentgenologic appearance of a well-established ulcerative colitis. The large bowel is markedly shortened and narrowed and there is stricture formation near the hepatic flexure. In addition, the colon is not distensible, and the mucosa shows the typical changes of the disease. In later roentgenograms (Fig. 1, *D* and *E*) there is gradual improvement in the appearance of the large bowel, and the narrowing and shortening are less marked. In the last series of roentgenograms the barium filled colon appears normal (Fig. 1*F*), but in the postevacuation study (Fig. 1, *G* and *H*) there is distortion and beading of the mucosal pattern with longitudinal furrowing, showing it to be still definitely abnormal. Thus, in our case, when the barium filled colon alone is used as a criterion, there appears to be complete roentgenologic remission of ulcerative colitis. But, if the appearance of the mucosal pattern as seen on the postevacuation roentgenogram is employed to judge remission, the unmistakable traces of the disease remain.

Shanks and Kerley[9] state that in ulcerative colitis, "*restitutio ad integrum*" takes place only if cure is obtained early in the disease, and Feldman[3] describes the changes as "permanent." Others[2,4,6,8] use terms such as "stenosed," "cicatricial," "fibrosis," and "scarring" to imply that the far advanced changes in ulcerative colitis are not reversible. However, it was pointed out by Bockus *et al.*[1] that occasionally a colon which is markedly stenosed due to advanced ulcerative colitis can be distended by barium enema during a clinical remission, and he illustrated his point by citing a case that showed apparent roentgenographic improvement during a remission.

During the past twenty years, occasional cases have been reported that showed improvement in the roentgenologic appearance of the colon, and, in some cases, the diseased colon apparently has become normal. Ricketts, Kirsner, and Palmer[7] reported complete roentgenologic recovery in 5 cases of ulcerative colitis, but the appearance of the mucosal pattern is not mentioned. Kirsner, Palmer, and Klotz[5] cited 4 cases of complete roentgenologic remission. In 1 case, the mucosal pattern is mentioned as abnormal during an exacerbation, but the reversibility of the mucosal abnormality later is only implied, and not definitely stated.

In reviewing the literature in all cases so far reported, the reversion to normal has been demonstrated only by the barium filled colon, and the mucosal pattern as seen in the postevacuation roentgenogram has not been shown. Since the mucosa is the primary site of involvement in ulcerative colitis, it would seem logical that the mucosal pattern should be demonstrated to be normal when attempting to establish a complete roentgenologic remission in ulcerative colitis.

SUMMARY AND CONCLUSION

A patient with far advanced ulcerative colitis that showed striking improvement both clinically and roentgenologically is reported. Attention is drawn to the fact that the roentgen appearance of the barium filled colon alone is a poor criterion for judging complete reversal of the process. A normal mucosal pattern should be demonstrated before complete roentgenologic remission can be claimed.

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Vol. 86, No. 5

PNEUMATOSIS OF THE INTESTINES

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PNEUMATOSIS or gas cysts of the intestine (pneumatosis intestinalis cystoides) is a descriptive name denoting an abnormal collection of gas in the walls of the gastrointestinal tract. Numerous case reports and reviews of this condition have appeared in the literature since its first description by Du Vernoi in 1738.

Pneumatosis of the intestine is not a disease entity. It is a symptom complex accompanying abdominal diseases of different etiology and occurring in every age group of patients (from three days to seventy years of age).

Our purpose is to report upon that group of pneumatosis seen in adult patients who show marked weight loss, malnutrition, or even emaciation, and to present the results of treatment. As pneumatosis is of rather rare occurrence, there are uncertainties about its etiology, course and treatment. The observations made on our patients may throw some light on its etiology and may indicate the treatment of choice.

REPORT OF CASES

CASE 1. J.J., a thirty-three year old male, was admitted as an emergency patient with the provisional diagnosis of perforated peptic ulcer. The diagnosis was based upon a past history of duodenal ulcer and the presence of a marked hydropneumoperitoneum on fluoroscopy.

For seven days prior to admission he had vomited after every meal and had noted painless increasing abdominal distention and splashing sounds emanating from his abdomen. During the last six months the patient had lost 6 kg. in weight.

Physical examination showed a markedly undernourished, dehydrated, but comfortable, patient. His height was 175 cm. and weight 52.5 kg. The abdomen was extremely distended, but soft. Shifting dullness was present. The rest of the physical examination was negative. Urinalysis and blood cell count were within normal limits.

FIG. 1. Case 1. Scout roentgenogram of the abdomen using horizontal rays in the supine position shows the extensive hydropneumoperitoneum.

Scout roentgenograms of the abdomen with horizontal rays (Fig. 1 and 3*A*) revealed extensive hydropneumoperitoneum, multiple cysts under the right diaphragm, interposition of intestinal loops between liver and diaphragm and absence of the liver shadow. On the scout roentgenogram using vertical rays in the supine position (Fig. 2), the liver shadow appeared in its normal position and the cysts were visible over the entire abdomen.

Because of the absence of clinical signs of an acute abdomen, a diagnosis of pyloric stenosis associated with pneumatosis intestinalis cystoides was made. Rehydration, gastric suction and parenteral alimentation were commenced.

In order to relieve the abdominal distention, 1,500 cc. of fluid was removed by abdominal tapping on the day of admission. This fluid, which was yellow colored, was sterile on bacteriologic examination. Guinea pig inoculation was negative for tubercle bacilli. After adequate rehydration the patient was fed on a concentrated high caloric soft diet.

Serum electrolytes and liver function tests were normal. Barium studies of the gastrointestinal tract showed a marked pyloric stenosis. The colon was normal; however, the

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FIG. 2. Case 1. Scout roentgenogram using vertical rays in the supine position shows multiple cysts scattered throughout the abdomen.

small intestine was noted to be interposed between the liver and diaphragm. Repeated scout roentgenograms showed a gradually diminishing pneumoperitoneum, disappearance of free fluid in the peritoneal cavity and some reduction in the number of cysts (Fig. 3, *A*, *B* and *C*).

The patient was prepared for surgery by repeated blood transfusions, amigen, glucose and electrolyte infusions. Operation was carried out on the twenty-third day of hospitalization. The day before operation a small pneumoperitoneum and numerous cysts were still visualized on roentgenograms.

Before opening the peritoneal cavity, peritoneal air was obtained for gas analysis through a cannula by means of a vacuum apparatus. The stomach was found to be dilated and have thickened walls, and an almost complete pyloric obstruction due to a chronic duodenal ulcer was seen. Five thousand cubic centimeters of yellowish ascitic fluid were removed from the peritoneal cavity.

A segment of the distal ileum, about 80 cm. long, was covered by numerous gas cysts, 1–3 cm. in diameter, arranged in clusters on the mesenteric and the antimesenteric borders of the bowel. They were round, of elastic consistency, and translucent. Some of them adhered to each other and to the intestinal wall. Others were single, attached to the free end of a thin avascular pedicle, often bluish in color, and contained small amounts of fluid (Fig. 4). Segments of normal bowel were observed between the masses of gas cysts, as well as fine white scars and coarse adhesions between the loops. The mesentery was edematous, and signs of mild inflammatory reaction along the affected segments were noted. The mesenteric lymph nodes were enlarged. The gas in the cysts could not be pressed from one cyst into another or into the intestinal lumen. On pressure the cysts burst. There was no other intra-

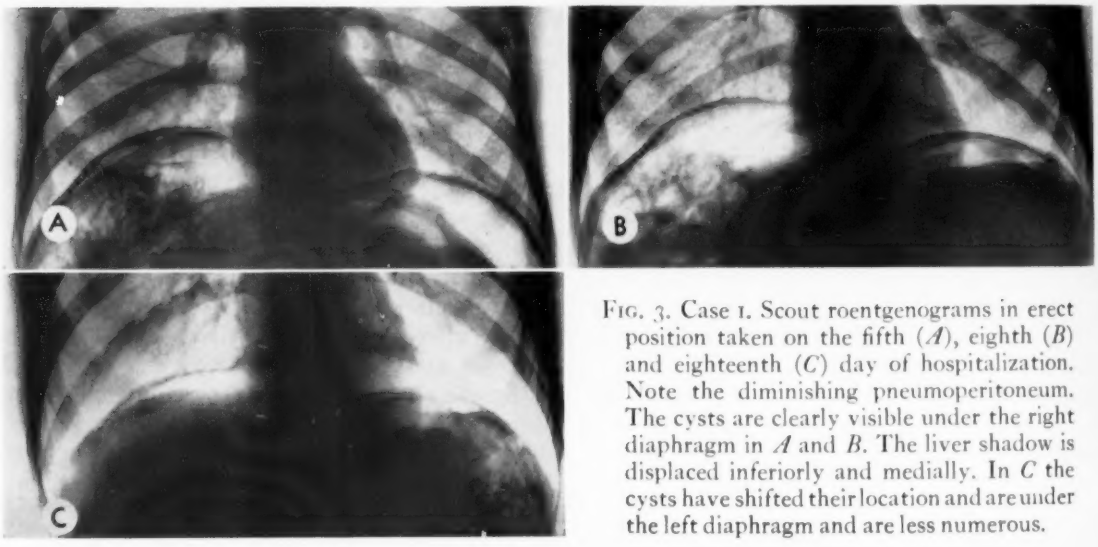

FIG. 3. Case 1. Scout roentgenograms in erect position taken on the fifth (*A*), eighth (*B*) and eighteenth (*C*) day of hospitalization. Note the diminishing pneumoperitoneum. The cysts are clearly visible under the right diaphragm in *A* and *B*. The liver shadow is displaced inferiorly and medially. In *C* the cysts have shifted their location and are under the left diaphragm and are less numerous.

abdominal pathology. One large gas cyst was excised intact so that a gas analysis could be obtained. A transmesocolic gastrojejunostomy and a bilateral vagectomy were performed. The analysis of the gas obtained from the peritoneal cavity and from the resected cyst showed 79 volume per cent nitrogen, 19 volume per cent oxygen and about 1 volume per cent CO_2.

The postoperative course was uneventful. A scout roentgenogram of the abdomen taken on the eighth postoperative day showed complete disappearance of the cysts (Fig. 5).

On the twelfth postoperative day the patient was discharged. He gained 10 kg. in weight during the first seven postoperative weeks and 22 kg. in the six months following his operation. Roentgenologic re-examination six, twelve, eighteen, twenty-four and forty months after surgery did not disclose any cysts or pneumoperitoneum.

The patient is at present in excellent health.

Comment. This patient was in an extremely poor nutritional state due to severe pyloric obstruction. He presented the typical roentgen findings of pneumatosis intestinalis cystoides with marked ascites of the reactive exudative type. The pronounced hydropneumoperitoneum was in sharp contrast to the mild clinical picture. No urgent surgery was done. On conservative treatment the pneumoperitoneum receded gradually and the cysts became less numerous. Eight days after the gastrojejunostomy and vagectomy, all the cysts and the pneumoperitoneum disappeared and did not recur during a three year follow-up. To our knowledge this is the shortest reported interval for the postoperative disappearance of gas cysts.

Fig. 4. Case 1. Appearance of a cyst-bearing loop at operation. Note the long pedicles of the cysts.

Fig. 5. Case 1. Scout roentgenogram on the eighth postoperative day. No cysts are visible.

Case 11. J.M., a seventy year old male, was admitted as an emergency patient because of abdominal pain, intractable vomiting, a distended abdomen and severe dehydration. The past history revealed a roentgenologically proved duodenal ulcer of twenty years' duration.

On physical examination the patient was extremely dehydrated and emaciated. His weight was 46 kg. and height 174 cm. Definite signs of avitaminosis were present in the angles of the mouth, buccal mucosa and on the skin of the forearms and the hands. Pitting edema of the lower extremities was present.

Scout roentgenograms of the abdomen (Fig. 6) showed pneumoperitoneum and the other characteristic findings of pneumatosis intestinalis cystoides. A diagnosis of pyloric obstruction, pneumatosis intestinalis cystoides and severe malnutrition was made. Rehydration, gastric suction and parenteral alimentation were started.

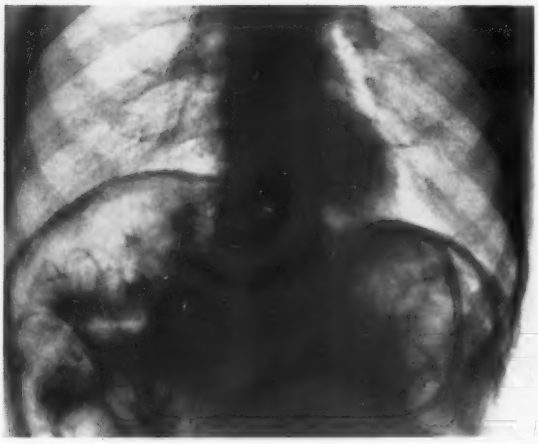

FIG. 6. Case II. Roentgenogram in the erect position shows an appearance similar to that seen in Case I (Fig 3*A*).

Laboratory examinations including urinalysis, blood cell count, liver function tests and serum electrolytes were within normal limits.

Roentgen examination on the fourth day of admission showed severe pyloric obstruction and rounded small filling defects in the lumen of the small intestine. Loops of small intestine were interposed between the liver and the diaphragm. These loops did not change their location with change of the patient's position. The transit of barium in the small intestine was markedly slow. A barium enema study was normal. Laminagraphy of the right upper abdomen (Fig. 7) showed adhesions between the walls of the intestine and the diaphragm.

FIG. 7. Case II. Laminagram of right upper abdomen. Note the large cyst below the diaphragm laterally and the adherence of cysts and intestine to diaphragm.

FIG. 8. Case II. Appearance of the cyst-bearing, doubly-rotated loop at laparotomy.

Preparations for surgery included blood transfusions, amigen and electrolyte infusions and high doses of vitamins. The patient was operated on seven days after admission. Laparotomy revealed a distended, thick-walled stomach and almost complete pyloric obstruction due to a duodenal ulcer. The distal part of the jejunum, with numerous large gas cysts attached, was interposed between the liver and the right diaphragm. There were numerous adhesions between the loops and the right diaphragm. This segment was doubly rotated on the axis of its mesentery (Fig. 8), the bowel wall was congested, the mesenteric vessels engorged, but no thrombotic process was evident. The segment of bowel proved to be viable after relieving the torsion. Another segment of small bowel, 130 cm. in length and extending to within 20 cm. of the ileocecal valve, was covered by multiple subserosal cysts varying in size from 1 to 5 cm. in diameter. They were thin-walled, translucent and round, and appeared on the mesenteric and antimesenteric borders of the bowel. They were either packed closely together and attached to each other and to the intestinal wall by a broad base, or they were single on the free end of an avascular pedicle. Short segments of normal small bowel were scattered between the masses of gas cysts. Signs of subacute and

chronic inflammation, with fine multiple adhesions and delicate white scar formations, were present between the wall of the intestines and the mesentery.

A transmesocolic gastrojejunostomy was performed and a large cyst was excised for gas analysis. The adhesions between the small intestinal loops and right diaphragm were divided

The postoperative course was uneventful. The signs of avitaminosis disappeared gradually. The patient gained 6 kg. in five weeks, and most of the gas cysts disappeared in that period as indicated by repeated flat roentgenograms of the abdomen. Six months postoperatively no cysts were visible roentgenographically. The result of the gas analysis of the cyst was the same as in Case 1.

The patient was discharged thirty-eight days postoperatively and followed-up for two years.

Repeat roentgenologic examinations revealed no evidence of the cysts.

Comment. This patient was emaciated due to long standing pyloric obstruction. The preoperative diagnosis was pyloric stenosis associated with pneumatosis intestinalis cystoides. No urgent surgery was undertaken. Elective gastrojejunostomy was performed after adequate preparation. The patient made an excellent recovery and the cysts gradually disappeared, the last of them six months after operation. Today, two years postoperatively, his general condition is satisfactory.

CASE III. F.S., a fifty-four year old male, was admitted as an emergency patient because of severe vomiting and vague abdominal pains. He had been vomiting after meals during the three months prior to admission. The past history revealed a duodenal ulcer (proved by roentgen examination) of sixteen years' duration, two episodes of hematemeses (six years and four years prior to admission), 8 kg. loss of weight and fainting spells during the last three months.

On admission the patient appeared markedly undernourished and dehydrated. His skin was atrophic and scaly. The abdomen was somewhat distended but no signs of peritoneal irritation were detectable.

Scout roentgenograms of the abdomen in erect position showed a moderate pneumoperitoneum. An unusual finding was a 2–5 mm. wide strip of air shadow outlining the stomach. This air strip was present on both the lesser and greater curvature as well as in the anterior wall of the stomach. Additional roentgenograms showed the air strip unchanged in its relation to the cavity of the stomach and its wall (Fig. 9, *A*, *B* and *C*), thus proving that it was trapped between the layers of the gastric wall. The stomach was markedly dilated and contained a large quantity of fluid.

The diagnosis of pyloric stenosis with gastric pneumatosis was made. No urgent surgery was deemed necessary. The patient was accordingly admitted for elective surgery after preparation by conservative treatment.

Urinalysis, blood cell count and liver function tests were all normal. The blood urea nitrogen was elevated due to extrarenal uremia explained by marked dehydration. There was severe electrolyte imbalance—hypopotassemia, hyponatremia, hypochloremia and metabolic alkalosis. This electrolyte imblance was completely corrected by the seventh day after admission by repeated blood transfusions and parenteral electrolytes. On the eighth day of hospitalization no free air was seen in the peritoneal cavity and only small bubbles of air could be detected in the stomach wall.

At laparotomy on the twelfth day after admission the stomach was found to be greatly dilated with thickened walls. Crepitation was felt on palpation of the stomach wall. Tight pyloric obstruction was present. No perforation was seen. No cysts were found on the small or large intestines, neither were cysts seen on the peritoneal surface of the stomach. Gastrojejunostomy with vagectomy and a feeding jejunostomy were performed.

The postoperative course was uneventful. The temporary feeding jejunostomy tube was withdrawn on the seventh postoperative day and the patient was discharged on the twelfth postoperative day, having gained about 4 kg. during his hospitalization of twenty-four days. At re-examination one year after surgery, the patient was in excellent condition. There was no air visible in the stomach wall.

Comment. This patient was admitted in a markedly undernourished condition due to prolonged vomiting caused by pyloric stenosis. The roentgen examination revealed the unique appearance of "gastric pneumatosis" in addition to a pneumo-

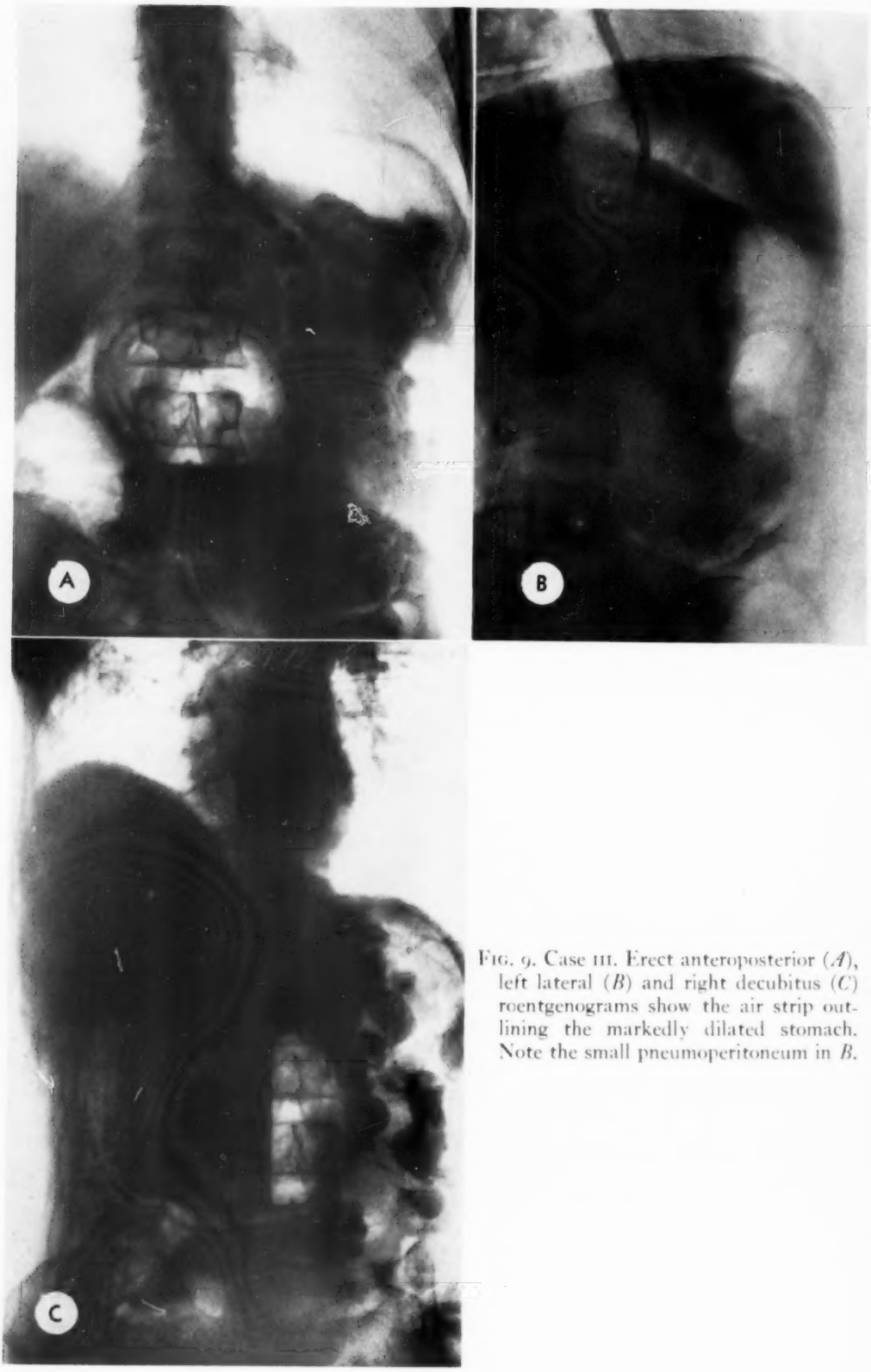

Fig. 9. Case III. Erect anteroposterior (*A*), left lateral (*B*) and right decubitus (*C*) roentgenograms show the air strip outlining the markedly dilated stomach. Note the small pneumoperitoneum in *B*.

peritoneum. No other signs of pneumatosis of the intestines were seen. After elective gastroenterostomy and vagectomy the patient recovered completely.

To our knowledge this is the first case in which this type of gastric pneumatosis has been roentgenologically demonstrated.

Case IV. U.A., a forty-four year old male, was admitted as an emergency patient because of severe vomiting and vague abdominal pains. His past history revealed that during the previous nine years he had had five episodes of sudden marked abdominal distention with vomiting and constipation. These episodes occurred at intervals of about one to two years and always after extreme physical and mental strain. On each occasion a pneumoperitoneum was diagnosed clinically and roentgenologically. Seven years previously, during his second episode, laparotomy was done (in Romania) but nothing abnormal was found. The pneumoperitoneum subsided on each occasion upon rest and nutritional improvement about six to seven weeks after its appearance.

The present episode of sudden abdominal distention occurred fourteen days after his arrival in this country from Romania. He had had quite a lot of trouble connected with his emigration, his arrival and with settling down anew.

On admission his abdomen was extremely distended, but no signs of an acute inflammatory or obstructive abdominal condition could be detected. His nutritional condition was rather poor. The rest of the physical examination was noncontributory.

Roentgen examination revealed a marked pneumoperitoneum with the presence of a delicate, curved linear shadow below the right diaphragm, resembling the wall of a gas cyst.

A diagnosis of recurrent pneumoperitoneum, probably due to pneumatosis intestinalis cystoides, was made. Urgent surgery was not indicated. The patient responded well to conservative treatment and was discharged for follow-up in the outpatient department. A barium meal study showed a normal esophagus, stomach, duodenum and small intestine. No stenotic lesion was seen in any part of the gastrointestinal tract. The pneumoperitoneum disappeared three weeks after admission.

Follow-up examination one year after discharge showed a well-nourished man with no complaints. Flat roentgenograms of the abdomen showed a rather unusual quantity of air in the stomach and small intestine without fluid levels. No pneumoperitoneum was seen.

Comment. This undernourished patient under emotional stress due to his recent arrival in the country was admitted with a marked pneumoperitoneum. No signs or symptoms of peritoneal inflammation were present and, therefore, the abdomen was not explored. No ulcerative and/or stenotic lesion was found on subsequent roentgenologic examination of the gastrointestinal tract. The pneumoperitoneum disappeared three weeks after admission upon improvement of his physical condition.

Re-examination a year later showed a well-nourished happy man.

DISCUSSION

The 4 above-described cases have many common features, but show some different findings as well.

The most evident common findings in all cases were pneumoperitoneum, malnutrition and vomiting, on one hand, and absence of signs of an acute abdomen, on the other. The pneumoperitoneum disappeared gradually on conservative treatment in all patients. In our opinion the disappearance of the pneumoperitoneum was due to the improved nutritional state.

The severe malnutrition in the first 3 cases is well explained by the long-standing marked pyloric obstruction. In the fourth patient the malnutrition was probably due to his emotional stress and physical strain. Reports dealing with pneumatosis intestinalis cystoides in association with a stenotic lesion of the gastrointestinal tract nearly always mention that the patients were in a poor nutritional state.[1,2,5,9,11,13,14,16,17,18] In a previous report from this hospital, a case of pneumatosis intestinalis cystoides with severe malnutrition was described.[6,15] We believe, therefore, that pneumatosis intestinalis cystoides of the described type is most probably due to malnutrition.

As we were reasonably certain that im-

mediate surgical intervention was not indicated, it was decided to prepare the patients for elective surgery by treatment aimed primarily at improving their nutritional state. All cases responded well to this treatment; the pneumoperitoneum diminished markedly or disappeared, the number of cysts became smaller and the gas in the stomach wall nearly disappeared. The complete disappearance of the cysts after gastroenterostomy also supports our opinion. Gastroenterostomy, by allowing normal food intake, removes the cause of malnutrition which, in turn, results in the disappearance of the cysts. After functional correction of pyloric obstruction, this has been observed by other authors as well.[1,2,4,5,7–10,17]

Follow-up examinations of the 3 operated cases showed that the patients remained in good nutritional state and neither the pneumoperitoneum nor the cysts recurred during a period ranging from one to three years. We have had under observation for four years another patient, severely malnourished, with pyloric obstruction and pneumatosis intestinalis cystoides who refused surgery.[15] The typical findings of pneumatosis intestinalis cystoides were present on repeat examinations, proving that this process persists as long as malnutrition is not corrected.

The rewarding results of the treatment given to our patients show that preoperative conservative therapy is indicated and that elective surgery should consist of relieving the obstruction. The cyst-bearing loops may be left alone. In Case II, volvulus of the cyst-bearing small intestinal loops and adhesions between these loops and the diaphragm were present. Though the involved loops were edematous and covered by numerous large cysts, no resection was carried out. The presence of even very numerous and/or large cysts is not an indication for resection.

The diagnosis of pneumatosis intestinalis cystoides should be made without difficulty by roentgenologic examination in typical cases. The clinician, however, can suspect the diagnosis when a pneumoperitoneum is detected in a malnourished patient, who is not acutely ill. In addition, the history of a long-standing duodenal ulcer makes the diagnosis probable. Flat roentgenograms of the abdomen in erect and supine positions show the pneumoperitoneum, the interposition of the small intestine between the liver and the diaphragm and the delicate ring shadows representing the cysts in the typical cases. In Case III, the roentgenographic finding of air in the gastric wall, without the usual appearance of cysts, was surprising. The pneumoperitoneum and the known long-standing pyloric obstruction suggested the diagnosis of pneumatosis intestinalis cystoides. We believe, therefore, that the gas in the gastric wall is equivalent to cysts on the intestines seen in other cases. To our knowledge this is the first case in which pneumatosis of the stomach presented itself in the form of a continuous intramural layer of air without cyst formation. Isolated cases of cysts on the stomach with pneumatosis of the small intestine, however, have been described.[2,7]

Case IV had only two signs in common with the first 3—pneumoperitoneum and malnutrition. Such isolated cases have been reported as idiopathic pneumoperitoneum. The pneumoperitoneum in this patient disappeared spontaneously upon rest and improvement of his nutritional state, and reappeared again with malnutrition. We believe, therefore, that this case has some relationship to pneumatosis intestinalis cystoides.[6]

As stressed above, this paper deals with pneumatosis of the benign type seen in adults only. We believe that this type of pneumatosis is due to malnutrition. This conception is supported by the following facts: (1) pneumatosis of this type is always associated with malnutrition; (2) it disappears shortly after correction of the deficient nutritional state; and (3) it perseveres with persistent malnutrition.

SUMMARY

1. Two typical, one atypical and one probable case of pneumatosis intestinalis cystoides are described. All are associated with malnutrition.

2. After correcting the malnutrition by conservative and surgical treatment, the pneumatosis disappeared.

3. The cyst-bearing parts of the gastrointestinal tract do not require resection even in cases with extensive pneumatosis because the cysts disappear spontaneously.

4. In one case the cysts completely disappeared eight days after surgery—the shortest time so far observed.

5. A hitherto unreported type of intramural gastric pneumatosis without cyst formation is described.

6. The so-called idiopathic pneumoperitoneum has probably some relationship to pneumatosis intestinalis cystoides.

7. We believe that malnutrition is the main etiologic factor of this type of pneumatosis intestinalis cystoides.

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GAS IN THE PORTAL VENOUS SYSTEM*

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THE presence of gas in the intrahepatic branches of the portal vein during life is a little known and seldom recognized condition of grave prognostic significance. It is readily detectable roentgenologically, the intravascular gas producing a branching linear pattern. As will be shown, this pattern can be distinguished from that of gas in the biliary tract.

On reviewing the literature between 1922 and 1960, we were able to find only two articles dealing with this subject. The first of these, by Wolfe and Evans[6] in 1955, reported the condition in 6 infants, ranging in age from fifteen days to six months, who were suffering from a variety of diseases. The second, by Susman and Senturia[4] in 1960, included an additional patient, an adult with mesenteric infarction, in whom the gas was detectable roentgenographically and confirmed at operation.

We have encountered 4 patients with gas in the portal system at the Cincinnati General and Children's Hospitals during the past three years. In 3, the classic roentgen features were evident. In the fourth, roentgenograms were not available, the gas being found at autopsy. In one of the cases (Case 1), the nature of the gas and its pathogenesis have been established with reasonable certainty.

REPORT OF CASES

CASE 1. W.W., a sixty-three year old white man, entered the Cincinnati General Hospital on July 21, 1960, with a six hour history of severe abdominal pain, distention, and diarrhea. He had been on peptic ulcer management because of a prior history of postprandial pain and two episodes of tarry stools, the last of which had occurred six months earlier.

Physical examination revealed a temperature of 96.5° F. and a pulse of 130. Positive findings were limited to the abdomen, which was distended and tympanitic. Following gastric aspiration of three liters of thick foul brown fluid and air, the distention subsided and the abdomen became soft and nontender. On rectal examination a large amount of bright red watery stool was noted. Sigmoidoscopy was normal.

FIG. 1. Case 1. Detail view of the liver area on admission showing a few branching linear gas shadows (arrow) near the periphery of the right lobe.

Laboratory studies revealed a hemoglobin of 12 gm. and a white blood cell count of 12,300 per cu. mm., with 80 per cent neutrophils. The blood urea nitrogen, CO_2 combining power, and serum amylase were normal. The blood chloride was slightly decreased, and the blood sugar was 253 mg. per cent. The clotting time was slightly prolonged but the bleeding and prothrombin times and platelet count were normal.

Recumbent and upright roentgenograms of the abdomen showed marked distention of the stomach and moderate distention of the small bowel with multiple gas-fluid levels. Within the confines of the liver shadow were several small branching linear gas shadows (Fig. 1). It was felt that they represented gas in the portal system, probably secondary to necrosis of the

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bowel. Roentgenograms made the following day no longer showed these branching shadows. Barium enema examination was normal.

Exploratory laparotomy was performed two days after admission. Marked distention of the small bowel hampered the procedure. One member of the surgical team thought that a 180° volvulus of the small bowel was present, but another disagreed. A ten inch segment of partly gangrenous ileum, lying about four feet proximal to the ileocecal valve, was resected.

Postoperatively, the patient improved considerably. Although the diarrhea persisted, it was no longer bloody. Five days after the operation the patient suddenly went into shock and complained of severe abdominal pain. Portable roentgenograms of the abdomen revealed free intraperitoneal air, small bowel distention, and two segments of edematous bowel in the right lower quadrant. No gas was visible in the hepatic vessels. Stool guaiac test was 4 plus. Blood culture revealed abundant *Aerobacter aerogenes*. The patient remained in shock, became markedly oliguric and acidotic, and died the following day. The body was taken immediately to the radiology department where an abdominal roentgenogram was obtained (Fig. 2, *A* and *B*). This revealed extensive gas in the portal veins and in several mesenteric veins. In addition, there was considerable gas in the extraperitoneal space adjacent to the bladder, in the wall of several segments of intestine, and in the abdominal wall.

At autopsy marked crepitation of the abdominal and thoracic parietes, neck, and scalp was noted. Free peritoneal gas was found. Gas was visible in the mesenteric, portal, hepatic, and jugular veins, in vessels at the base of the brain, in the right atrium, and in the abdominal wall, extraperitoneal space, serosa of the bowel, and mesentery. Before dissection was carried out, gas was withdrawn from the portal and jugular veins and the right atrium by a vacuum syringe. It was felt that some contamination of the syringes with room air probably occurred.

The peritoneal cavity contained 2,500 cc. of cloudy light brown fluid. The mesenteric vessels were normally patent. Enteric contents leaked from the site of anastomosis. The intestines, especially the distal colon, showed intense congestion, hemorrhage, and areas of necrosis. The stomach showed similar findings and, in addition, a large chronic active prepyloric ulcer which had penetrated into the pancreas. The gallbladder and bile ducts were normal. All vessels leading to and from the liver were ligated prior to its removal. A roentgenogram

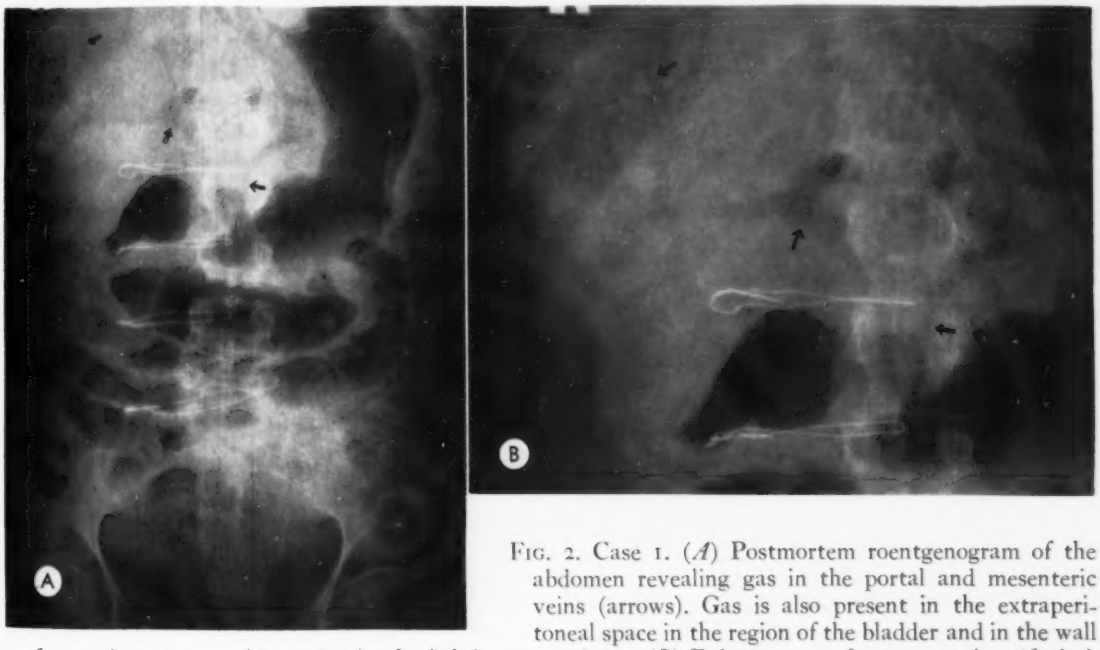

Fig. 2. Case 1. (*A*) Postmortem roentgenogram of the abdomen revealing gas in the portal and mesenteric veins (arrows). Gas is also present in the extraperitoneal space in the region of the bladder and in the wall of several segments of intestine in the left lower quadrant. (*B*) Enlargement of upper portion of *A*. A large mesenteric vein (lower arrow), the portal vein (middle arrow), and its intrahepatic branches (upper arrow) are seen.

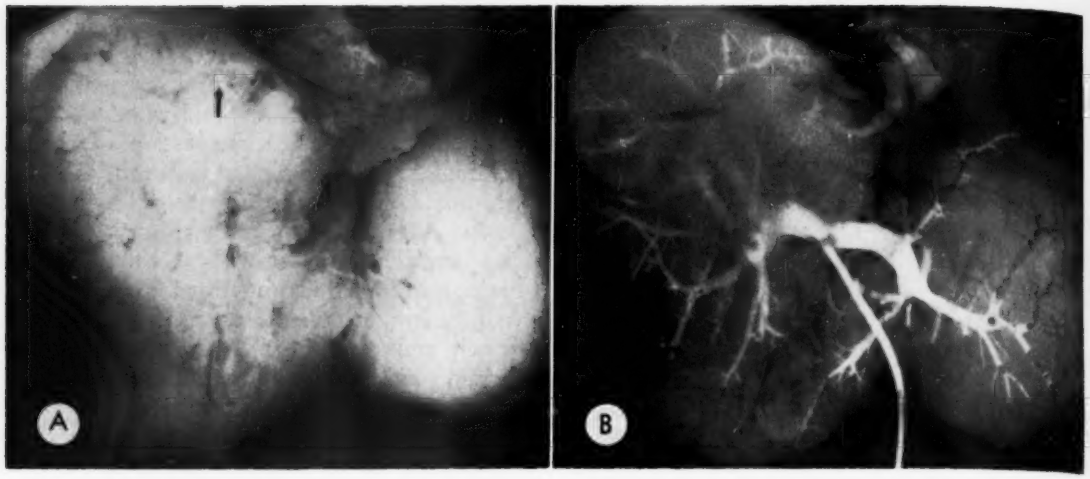

FIG. 3. Case I. (*A*) Roentgenogram of the liver removed after ligation of vessels at autopsy. Gas is seen within the portal veins and in at least one large hepatic vein (arrow). (*B*) Roentgenogram of the liver following the injection of barium into the portal system demonstrating the location of the gas. A later injection of the hepatic veins confirmed the location of the gas near the dome of the liver.

of this organ revealed considerable intravascular gas (Fig. 3*A*), some of which was withdrawn with an oiled needle and syringe. Barium was then injected into the main portal and hepatic veins, and a roentgenogram showed that most of the gas lay in the portal branches with a small amount in a large hepatic vein (Fig. 3*B*).

Microscopic sections of the colon, small bowel, and stomach showed hemorrhage, congestion, and necrosis. A large number of bacteria was visible in the sections. Several large spaces were seen in the submucosa of the stomach. Clusters of bacteria were present in the wall of the aorta and in small arteries and lymphatics in some of the viscera.

Cultures of the blood, lung, bowel, liver, and peritoneal fluid showed an abundant growth of *Aerobacter aerogenes*, interspersed with a few colonies of *Staphylococcus aureus* and *Streptococus alpha-hemolyticus*. The gas analyses yielded 24 per cent CO_2 in the gaseous phase in the liver, 28 per cent in the portal vein, 38 per cent in the jugular vein, and 35 per cent in the right atrium. The final pathologic diagnosis was acute hemorrhagic necrotizing gastroenterocolitis[2,5] and *Aerobacter aerogenes* septicemia.

Comment. A sixty-three year old man entered the hospital with acute abdominal symptoms and bloody diarrhea. Abdominal roentgenograms showed gas in the hepatic vessels and distention of the stomach and small bowel. Exploratory laparotomy revealed a necrotic segment of small bowel. Five days postoperatively he went into shock and died. A postmortem roentgenogram of the abdomen revealed extensive intravascular and interstitial gas. At autopsy, gas was present in the mesenteric, portal, and jugular veins and in the heart. The bowel showed acute hemorrhagic necrotizing gastroenterocolitis. Both antemortem and postmortem blood cultures grew *Aerobacter aerogenes*. Analysis of the gas revealed a high content of carbon dioxide.

CASE II. R.A., a thirty-five year old Negro male, was admitted to the Cincinnati General Hospital on June 21, 1958, comatose and in shock. He was a known diabetic and had taken his insulin the day prior to admission. While driving from Alabama with several companions, he became "car-sick" and ingested nothing but ice water and orange juice for twenty-four hours. On the following morning he could not be awakened and was brought to the hospital.

The temperature and pulse were normal but the respirations were 28 per minute. The blood pressure could not be obtained. Physical examination revealed anisocoria, absence of corneal reflexes, hypoactive tendon reflexes, and marked abdominal distention without audible bowel sounds.

The blood urea nitrogen was 84 mg. per cent and the sugar 854 mg. per cent. The CO_2, blood chlorides, and pH were extremely low. There was 4 plus glycosuria with moderate acetonuria. The stool guaiac test was negative.

Recumbent and decubitus roentgenograms of the abdomen (Fig. 4, *A* and *B*) revealed gaseous distention, severe in the small bowel and moderate in the ascending colon, with an occasional fluid level. Many slender, branching, linear gas densities were visible within the right side of the liver, increasing somewhat in caliber as they converged toward the hilus, but not reaching the size of a major vessel or duct. There was no gas in the gallbladder or extrahepatic biliary ducts. At first an internal biliary fistula was suspected, but subsequently a diagnosis of portal vein gas was made.

Despite supportive measures, including vigorous treatment of the diabetic acidosis, the patient expired eight hours after admission.

Autopsy was performed forty-eight hours post mortem, so a blood culture was not obtained. The small bowel was quite distended and its mucosa showed marked congestion but no evidence of ulceration or necrosis. No mention was made of intramural or intravascular gas, the prosector having had no knowledge of the roentgen findings. The stomach, colon, gallbladder, and biliary ducts showed no abnormality. The mesenteric vessels appeared normal. The liver was grossly normal and microscopically showed only mild congestion. With the exception of marked pulmonary edema, no other significant abnormalities were noted. The cause of death was listed as diabetic acidosis.

Comment. A diabetic entered the hospital in shock and acidosis and died eight hours later. Roentgenograms revealed intravascular gas in the liver and severe nonobstructive ileus. At autopsy the small bowel showed marked distention and mucosal congestion.

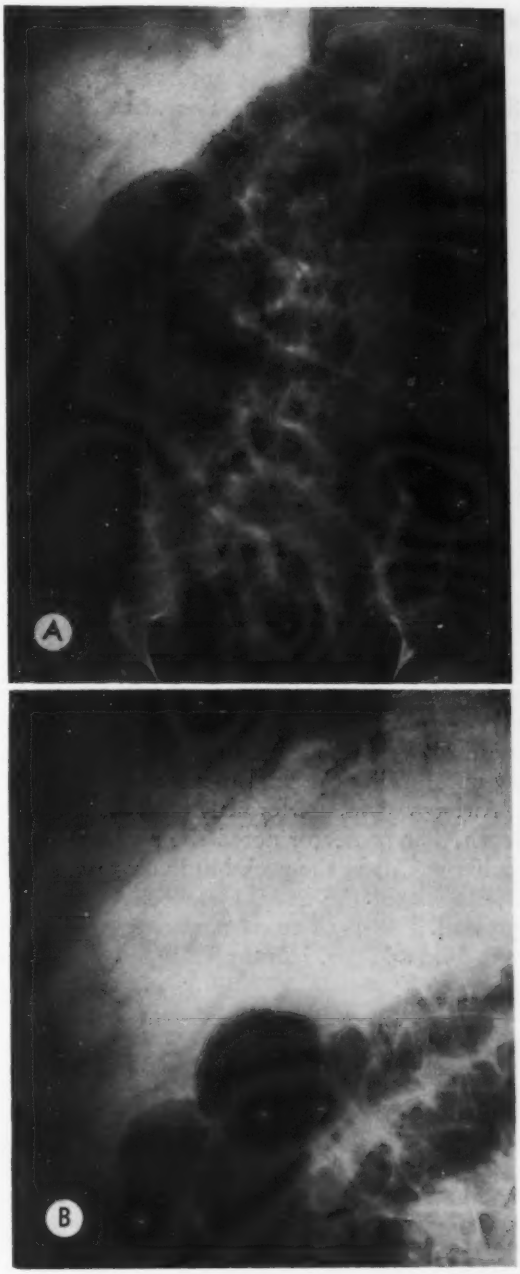

Fig. 4. Case II. (*A*) Recumbent roentgenogram revealing extensive gaseous distention of the intestines. Branching linear gas shadows are visible within the right side of the liver. (*B*) Detail view of the liver area.

Case III. J.A., a Negro female aged fifty-two, a diabetic of at least eleven years' duration, was admitted to the Cincinnati General Hospital on December 5, 1957, with a four day history of lower abdominal pain, diarrhea, and vomiting. She had eaten very little during the current illness but had taken 20 units of insulin per day. Bilateral supracondylar amputations of the lower extremities had been performed about ten years earlier. She had been blind for one year.

On admission the blood pressure was 215/105. The temperature was normal but the pulse was 104. She was drowsy but coherent. The

abdomen revealed hyperactive bowel sounds and diffuse tenderness.

The white blood cell count was 29,000 with 86 per cent neutrophils. The urine showed 2 plus sugar, 1 plus acetone, and 4 plus albumin. The blood urea nitrogen was 42 mg. per cent and the blood sugar 253 mg. per cent. The CO_2 and blood chlorides were slightly decreased. The stool was positive for occult blood.

Flat and decubitus roentgenograms of the abdomen revealed a few slightly dilated loops of small bowel. The following day the patient's general condition worsened. Abdominal roentgenograms now revealed marked small bowel distention with many fluid levels. Several thin linear gas shadows were visible in the right lower quadrant, apparently lying in intestinal wall or mesenteric vessels. Within the confines of the liver shadow, a few thin, faint, bifurcating, linear gas shadows were noted. Several small gas bubbles were present in the right flank (Fig. 5, *A* and *B*).

Although mesenteric infarction was suspected from the time of admission, the condition of the patient precluded operation, and she died forty hours after admission and four hours following the final roentgenograms.

Autopsy was performed nine hours after death. The prosector noted the presence of gas within vessels in the neck but made no mention of gas within the liver, mesenteric vessels, or gut wall. The small bowel was greatly distended with fluid and gas. Its mucosa appeared swollen and markedly congested. The superior mesenteric artery was occluded about 7 cm. from its origin by a dry firm thrombus. The colic branches arose proximal to the occlusion. The colon, liver, gallbladder, and biliary ducts appeared normal.

Microscopic examination of the small bowel revealed many areas of necrosis. The liver showed pericentral congestion and occasional cellular necrosis but no evidence of inflammation. Postmortem blood culture yielded *Proteus mirabilis*, *Proteus aerogenoids* and *Clostridium paraputrificans*.

Comment. This diabetic female had mild acidosis and acute abdominal symptoms. Intravascular gas in the liver, marked small bowel distention, and linear gas shadows lying either within mesenteric vessels or the intestinal wall were seen roentgenographically a few hours before death. Autopsy revealed gas within vessels in the neck, and small bowel necrosis secondary to mesenteric artery thrombosis. Postmortem blood culture yielded three different gas-producing organisms.

CASE IV. D.I., a one day old boy, entered Children's Hospital with evidence of esophageal atresia. Two days later this was corrected surgically. The subsequent course was stormy and he expired on the sixth postoperative day. At autopsy gas was found in all chambers of the

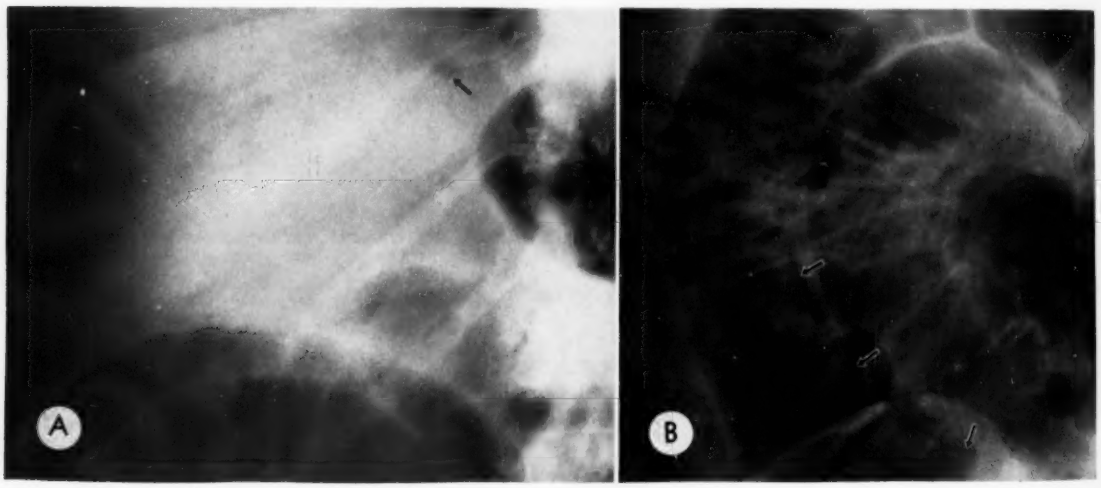

FIG. 5. Case III. (*A*) Enlarged view of liver area revealing a few bifurcating linear gas shadows (arrow). (*B*) Roentgenogram of right midabdomen showing a long linear gas shadow (arrows) lying either in the intestinal wall or in a mesenteric vessel. A small gas bubble is visible in the flank.

heart, in the inferior vena cava, in the portal vein, and in the intercostal arteries. The intestinal tract appeared normal. *Aerobacter aerogenes* was cultured from the blood. Microscopically, numerous bacterial colonies were seen in the liver. The final pathologic diagnosis was *Aerobacter* septicemia, pneumonia, and purulent mediastinitis.

DISCUSSION

In addition to the 4 cases described, one of us (B.F.) has seen 2 others with gas in the hepatic vessels. Unfortunately, this was over ten years ago and the patients' names have been lost, so that the roentgenograms and hospital records are not available for review. The first of these was a middle-aged man who entered the Dayton Veterans Hospital with signs and symptoms of small bowel obstruction. Roentgenograms of the abdomen revealed branching linear gas shadows within the liver and evidence of small bowel obstruction. At operation a closed loop small bowel obstruction with intestinal necrosis was found. The patient died postoperatively. Autopsy was not performed. The second case was that of a newborn infant who showed roentgen evidence of small bowel obstruction and gas within the hepatic vessels. The infant died without operation. Autopsy was not done. Because of limited information, these 2 cases were excluded from the present series.

Table 1 is a summary of the findings in the 11 recorded cases of intravascular gas in the liver, including the 4 in the present report. There were 7 infants and 4 adults. Nine were males. Only 2 were diabetic, both in the present series.

Antemortem blood culture was recorded in 2 cases, one being negative and the other showing *Aerobacter aerogenes*. In 9 of the 11 cases, postmortem blood cultures were performed and all were positive, *E. coli* being found in 6, *Aerobacter aerogenes* in 2, and *Clostridium paraputrificans* and two types of *Proteus* in 1. All of the *E. coli* cultures were encountered in Wolfe and Evans' series of infants, and both of the *Aerobacter* cultures were from our own series, one in an adult and the other in an infant.

Marked distention of the gut, shown roentgenographically or pathologically, was present in 10 of the cases. Although in every instance the intrahepatic gas was believed to lie within the portal system, in 5 cases it was actually visualized in the portal vein at autopsy. It was also recognized pathologically in mesenteric veins in 7 cases, in the inferior vena cava in 2, in a jugular vein in 2, in a hepatic vein in 1, in iliac and intercostal arteries in 1 each, in vessels at the base of the brain in 1, and in one or more cardiac chambers in 3. In addition, gas was demonstrated in the gut wall in 5 cases, pathologically in 4 and roentgenographically in 2.

All of the 8 patients who had roentgenograms showing intravascular gas died soon thereafter, only 2 surviving more than twenty-four hours. These died seven and eight days, respectively, after the first roentgen evidence of the gas.

The underlying diagnoses in the cases reported included, among others, 3 cases of erythroblastosis fetalis, 2 of mesenteric infarction, 2 of diabetes mellitus, 2 of necrotizing enterocolitis, and 1 each of imperforate anus, duodenal atresia, and esophageal atresia. Diarrhea occurred in 5 of the infants and 1 of the adults.

In searching among these cases for a common denominator which might explain the presence of gas in the vascular system, some interesting factors come to light. In the first place, all the organisms found on antemortem and postmortem blood cultures in these cases are potential gas-formers, especially in the presence of sugar. This raises the possibility of sepsis, at least in the portal circulation, as the cause of the gas. The well-known association of diabetes and gas-producing infection underscores the importance of the diabetes in Cases 11 and 111, and lends support to an infectious etiology of the intravascular gas.

The bacteria isolated in the blood cultures are all inhabitants of the normal intestinal tract. Necrosis of bowel wall, present in at least 4 of the 11 cases, certainly might predispose to their invasion

TABLE I

Author	Age	Sex	Diabetes	Blood Culture		Distention of Gut	Gas Other Than in Liver	Survival after Recognition of Gas	Other Diagnoses	Confirmation of Intravascular Gas
				Antemortem	Postmortem					
Wolfe and Evans[6]	17 da.	M	○	Negative	E. coli	+	Mesenteric veins; inferior vena cava; right atrium; wall of small and large intestine	15 hr.	Imperforate anus; pneumonia	Roentgenograms and autopsy
Wolfe and Evans	35 da.	M	○		E. coli	+	Mesenteric veins	7 da.	Duodenal atresia; postoperative intestinal obstruction; necrotizing enteritis	Roentgenograms and autopsy
Wolfe and Evans	15 da.	M	○		E. coli	+	Mesenteric veins; wall of large intestine	6 hr.	Erythroblastosis fetalis; cerebral vascular thrombosis; bronchopneumonia	Roentgenograms and autopsy
Wolfe and Evans	16 da.	M	○		E. coli	+		14 hr.	Erythroblastosis fetalis; bilateral renal cortical necrosis; pulmonary edema	Roentgenograms and autopsy
Wolfe and Evans	3 mo.	F	○		E. coli	+	Mesenteric veins; iliac arteries; wall of duodenum and jejunum		Diarrhea	Autopsy only
Wolfe and Evans	19 da.	M	○		E. coli	+	Mesenteric veins		Erythroblastosis fetalis; aspiration pneumonia; acute gastroenteritis	Autopsy only
Susman and Senturia[5]	77 yr.	M	○			+	Mesenteric veins	24 hr.	Superior mesenteric artery thrombosis	Roentgenograms and operation
Wiot and Felson Case I	63 yr.	M	○	Aerobacter aerogenes	Aerobacter aerogenes	+	Mesenteric, hepatic and jugular veins; right atrium; intestinal wall; extraperitoneal space; abdominal wall; mesentery; vessels at base of brain	8 da.	Acute necrotizing hemorrhagic enterocolitis; septicemia	Roentgenograms and autopsy
Wiot and Felson, Case II	35 yr.	M	+			+		8 hr.	Diabetic acidosis	Roentgenograms only
Wiot and Felson, Case III	52 yr.	F	+		Proteus; Clostridium	+	Cervical vessel (probably jugular vein); probably intestinal wall	4 hr.	Superior mesenteric artery thrombosis; diabetes	Roentgenograms and autopsy
Wiot and Felson, Case IV	6 da.	M	○		Aerobacter aerogenes	○	Intracardiac; inferior vena cava; intercostal arteries		Esophageal atresia; septicemia; pneumonia; purulent mediastinitis	Autopsy only

of the portal system. Unfortunately, the general unreliability of postmortem cultures lessens somewhat the validity of this argument. Even so, in Case 1 both antemortem and postmortem blood cultures grew abundant *Aerobacter aerogenes* and the postmortem tissues showed many bacterial colonies. *Aerobacter aerogenes* ferments various sugars to produce carbon dioxide and hydrogen.[1] Analysis of the gas collected at autopsy from the portal and jugular veins and right atrium of Case 1 showed high concentrations of carbon dioxide. The evidence for *Aerobacter* septicemia as the primary cause for the intravascular gas in this patient is, therefore, overwhelming. Case IV presents similar but less dramatic evidence of *Aerobacter* sepsis.

That septicemia from gas-producing organisms is the major factor in *all* cases of gas in the portal system cannot be implied from our present knowledge, although the uniformly fatal outcome in all the cases thus far reported suggests this possibility.

A theory is advanced by Susman and Senturia[4] that the gas from the distended intestine, under increased pressure, enters the mesenteric vessels through necrotic areas in the gut wall. This theory is supported by the recognition of "pneumatosis" in the gut wall in 5 of the 11 cases, by the severe gaseous distention of the intestine in all but one, and by the presence of enteric necrosis in at least 4 cases. Of course, these same factors would predispose to the passage of enteric organisms into the blood stream and thus also support the sepsis theory. Certainly, the theory of transmural migration of gas cannot explain the cases with intact intestinal mucosa.

It is our own belief that most, if not all, cases of intravascular gas in the liver result from gas-forming enteric organisms in the portal venous system, the intestinal distention, congestion, and necrosis probably playing an important role in the migration of the organisms through the intestinal wall.

The term "pneumatosis cystoides intestinalis," applied by Wolfe and Evans[6] to the intramural intestinal gas, is better avoided for the present, in view of its more specific connotation. We do not believe it is the instigating cause of the portal venous gas but consider it more likely the effect of a simultaneous intramural infection. The presence of gas within the hepatic veins, cardiac chambers, and peripheral veins indicates either extension of the gas, or of the organisms, through the portal capillaries.

The roentgen localization of the gas to the portal venous system and its differentiation from gas in the biliary system merit further discussion. Anatomically, the branches of the hepatic artery, portal vein, and bile duct lie together in the liver within a common sheath. Only their large extrahepatic portions show distinct differences in size, position, and direction. The hepatic veins, of course, show a completely different pattern as they enlarge and converge cephalad (Fig. 6, *A*, *B* and *C*).

There is no doubt that in all these cases the intrahepatic gas lies within the portal veins. It was actually seen within the portal system or its mesenteric tributaries in 9 of the cases, and it was clearly depicted roentgenographically in intrahepatic portal divisions in Case 1. Since the roentgen pattern of the hepatic veins bears no resemblance, the only other vascular system to be considered is that of the hepatic artery. We have been unable to find any case reports of gas within this system in the literature.

On the other hand, gas within the biliary system is more difficult to exclude. In this condition, when the gallbladder or extrahepatic bile ducts contain gas, as is usually the case, differentiation from intravascular gas is easy. If only the intrahepatic ducts show the gas, the problem becomes more troublesome. As pointed out by Susman and Senturia, however, most of the gas in the biliary tree, because of the caudal flow of bile, is found in the larger ducts. The reverse circumstances pertain in the case of gas in the portal venous system. Thus, gas seen predominantly in smaller peripheral branches is considered to lie in the portal

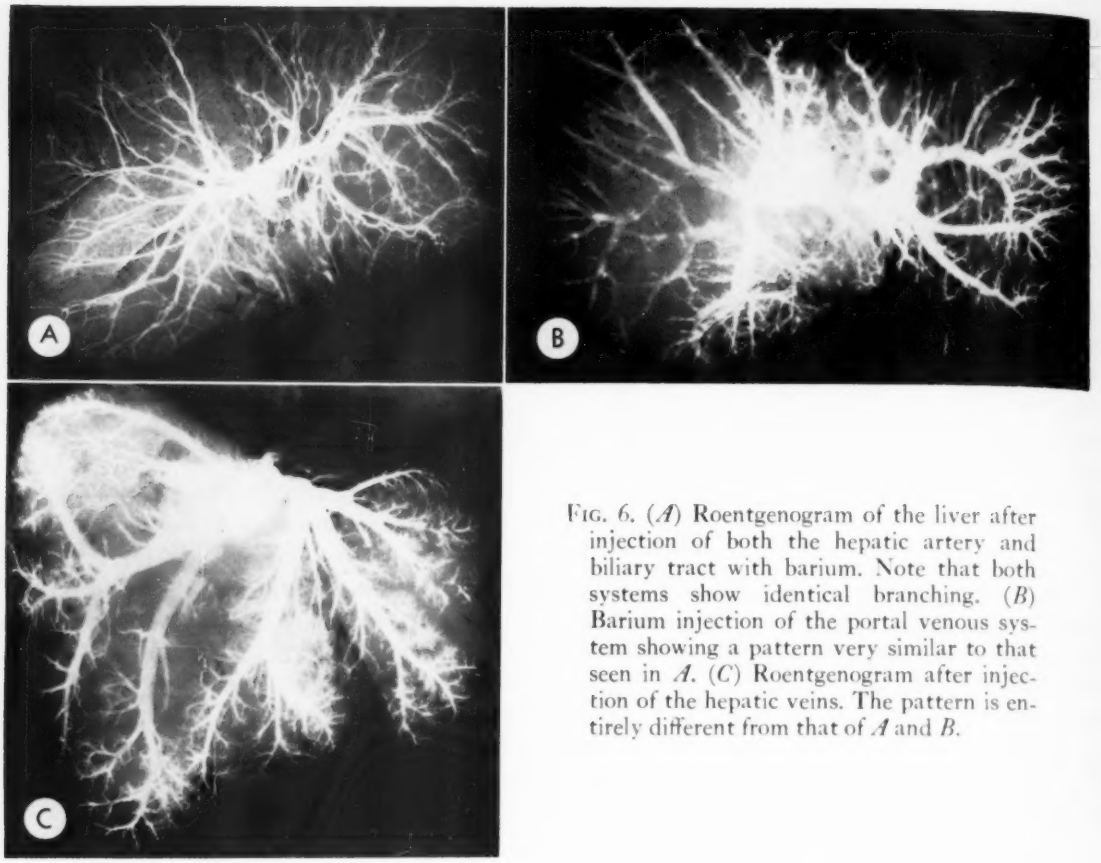

FIG. 6. (*A*) Roentgenogram of the liver after injection of both the hepatic artery and biliary tract with barium. Note that both systems show identical branching. (*B*) Barium injection of the portal venous system showing a pattern very similar to that seen in *A*. (*C*) Roentgenogram after injection of the hepatic veins. The pattern is entirely different from that of *A* and *B*.

vessels; if confined to larger, more caudal trunks, it is assumed to be in the biliary ducts. To date, this distinction has proved to be completely reliable. If doubt still exists, oral contrast medium may be used to attempt the delineation of an internal biliary fistula. On occasion, careful study of the roentgenogram will reveal gas in the mesenteric vessels or intestinal wall, as in 2 cases in the present series.

Some mention should be made of the occurrence of gas within the vascular system of the fetus, seen in cases of intrauterine death. Many explanations for this phenomenon have been offered,[3] but its pathogenesis has not been established. We do not believe that the mechanism is the same as that in the condition under discussion.

The presence of gas in the portal venous system is, at present, a sign of imminent death. All the patients have died, most within twenty-four hours. The longest survival was eight days from the time of its recognition. Whether the gas within the portal venous system is the actual cause of death or only an interesting terminal manifestation is unknown. For the present no specific therapy directed toward the intravascular gas itself is suggested. Aggressive treatment of the underlying disease and its complications (mesenteric infarction, sepsis, etc.) and, particularly, the use of those antibiotics effective against gas-forming organisms may result in the salvage of some cases in the future.

Of the 11 cases reported, 10 were seen in two institutions. This implies that the condition is considerably more common than the literature indicates, but that it is probably being overlooked. Careful examination of the hepatic region on the abdominal roentgenograms of all acutely ill patients

for these characteristic branching gas shadows will, we believe, result in the recognition of many more cases of this entity.

SUMMARY

Gas in the portal venous system presents a striking and diagnostic roentgenographic appearance which can be differentiated from gas in the biliary tree. Eleven cases, including 4 in the present series, have now been reported. The etiology has not been established, but evidence indicates that septicemia caused by gas-forming enteric organisms accounts for at least some cases. A high carbon dioxide content of the gas was established in one of the present cases. The frequency of distention, congestion, and necrosis of the intestine accompanying this condition strongly suggests an etiologic relationship. The prognosis is grave, all patients so far reported having died shortly after recognition of the intravascular gas.

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We wish to thank Drs. Merlin L. Cooper and Joseph T. Tamura for their advice concerning the bacteriologic aspects and Dr. Robert A. Kehoe and the Kettering Laboratory for performing the gas analyses in Case 1. We also are indebted to Drs. John J. Schlueter, William D. Suer, William Thomas, and Carol J. Amick for their help with the injection studies.

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THE INTRAMURAL CYSTIC DUCT REMNANT*

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IN TEN to fifteen per cent of patients with gallbladder disease, the symptoms are not relieved or they recur after cholecystectomy. One of the causes producing the so-called postcholecystectomy syndrome is the incomplete removal of the cystic duct, leaving behind a residual stump which can give rise to symptoms similar to those of gallbladder disease. This condition has been well described in the medical literature.[5,7]

This paper deals with the study of a less frequently recognized entity consisting of the presence of a cystic duct remnant of the intramural type. This results from incomplete removal of an anomalous portion of the cystic duct which is intimately bound to and hard to separate from the common bile duct.

ANATOMIC AND EMBRYOLOGIC CONSIDERATIONS

Normally, the cystic duct is from 1 to 1½ inches long and runs from the neck of the gallbladder to join the common hepatic duct at an acute angle, forming the common bile duct. Nevertheless, Flint[4] observes that the ducts do not unite immediately but are bound together in a connective sheet, and can be separated by careful dissection for a distance of from a few millimeters to several centimeters.

In reviewing the biliary tree of 45 cadavers, the author found that 25 had a normal angular junction of the cystic and hepatic ducts. In 10 cadavers the distal segment of the cystic duct ran parallel to the common duct and both were bound together by a fibrous union for a distance of less than 1 cm. In the remaining 10 cadavers studied, the two ducts were bound firmly together for lengths of 1 cm. to 1 inch; the bound segment of the cystic duct appeared to be embedded in the medial wall of the common duct but the lumens were separate (Fig. 1, *A* and *B*). A thin septum was found between the two ducts after careful dissection of the anterior wall. The gross appearance before dissection was that of a single duct.

This is the intramural variant of the cystic duct which occurred in 22 per cent of the cadavers studied. Similar findings were reported in 1929 by Beaver,[1] who found 26 per cent of 57 cadavers having a variation of the cystic duct similar to the one described here, which he called the "short parallel type."

This variation corresponds to what Boyden[2,3] calls "septate ductus choledochus" in classifying the developmental anomalies of the extrahepatic biliary tract. Embryologically it can be explained as produced by a double vacuolization of a segment of the common duct during the solid stage of development.

In the early embryonic stage the biliary ducts appear as hollow structures which later become solid as parietal cells migrate into the lumens. These solid epithelial structures then undergo a process of vacuolization, which restores the lumen. Almost always a single row of vacuoles appears, but in some instances a double row develops and gives rise to the formation of a septate common bile duct which will have an intramural segment of the cystic duct within its medial wall.

CLINICAL CONSIDERATIONS

An intramural remnant of the cystic duct may produce no symptoms after cholecystectomy, but it can be the site of pathologic changes such as those found in cases with remnants of the gallbladder or of the

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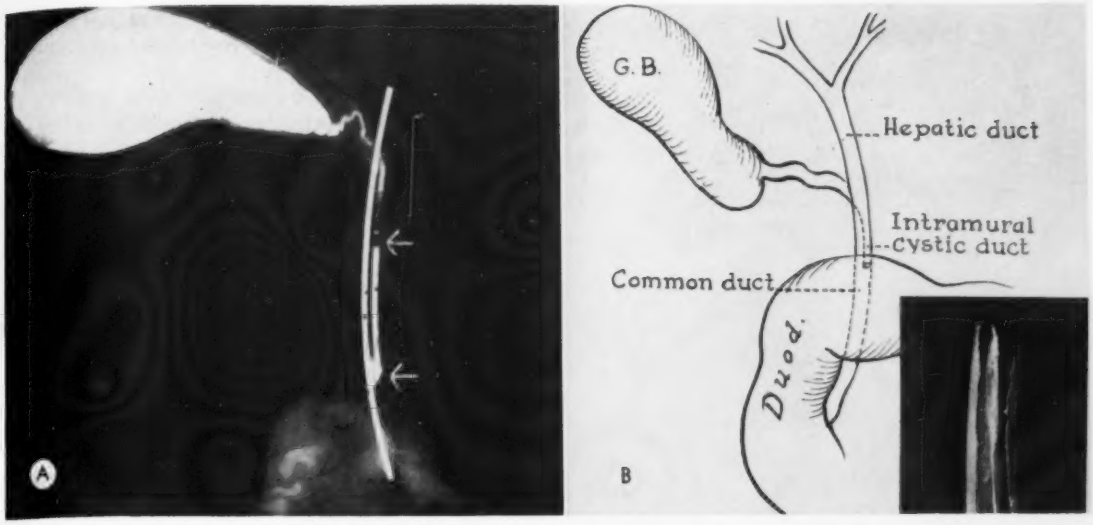

FIG. 1. (*A*) Anatomic specimen obtained from a cadaver and filled with barium. Thin polyethylene tubes were placed within the common and cystic ducts. A long intramural segment of the cystic duct measuring 1 inch in length was found running parallel to the medial aspect of the common duct (between arrows). (*B*)Diagram showing a normal biliary tree with an intramural segment of the cystic duct. The insert shows a segment of the anatomic specimen in *A* demonstrating the double lumen found at the distal portion of the common duct. The anterior wall of the duct has been dissected away. The septum separating the intramural segment of the cystic duct from the lumen of the common duct is clearly seen.

cystic duct. The symptoms may be indistinguishable from those of gallbladder disease. The need for a well-documented series of such cases is apparent in order to draw definite clinical conclusions.

ROENTGEN FEATURES

In reviewing a limited number of postcholecystectomy patients (55 cases) with studies of the biliary tree, two typical examples of an intramural cystic duct remnant were found. These will help to demonstrate the roentgen features of the condition described here. A large series will be published in the future.

The remnant of the cystic duct has been found only in the medial aspect of the common duct to date. It appears as a regular, clearly outlined segment of the distal cystic duct coursing parallel to the common duct and alongside its medial wall. The dividing septum between the remnant and the common duct can sometimes be visualized as a thin radiolucent line (Fig. 2). Nevertheless, no differentiation can be made between the two ducts at surgery.

The shadow of the intramural segment of the cystic duct may be superimposed on that of the common duct and thus may be mistaken for a normal variation of the opening of the cystic into the common bile duct (Fig. 3, *A* and *B*). These anomalies of the junction between the cystic and common ducts occur in about 10 per cent of cases,[8] and the cystic duct may open into the medial, anterior or posterior wall of the common duct.

CONCLUSION

An intramural segment of the distal portion of the cystic duct occurs more frequently than other anomalies of this portion of the extrahepatic biliary tree. Its incidence in the cadavers studied was high but apparently it does not contribute in any way to the production of gallbladder disease in the average individual. When cholecystectomy is performed and this anomaly is present, its subsequent behavior is similar to that of a cystic stump; and, if the remnant is long, dilatation ensues due to the difficulty in emptying. It may also be

FIG. 2. T-tube cholangiogram of an elderly woman who had persisting symptoms after cholecystectomy. A second operation revealed no abnormal findings. Symptoms continued after removal of the T-tube. The intramural remnant of the cystic duct is seen in its typical medial location (arrows). Comparison with previous cholangiograms showed a progressive dilatation of the remnant, which was originally mistaken for a fistulous formation.

associated with the formation of stones, which could produce symptoms.

If this anomaly is present, postcholecystectomy studies of the biliary tree by means of T-tube or intravenous cholangiography will often demonstrate it. Serial studies may also reveal progressive dilatation or the presence of residual stones. The radiologist should be aware of this anatomic variation in order to differentiate it from other conditions, such as a common duct fistula which was suspected in the case shown in Figure 2.

Operative cholangiography will show

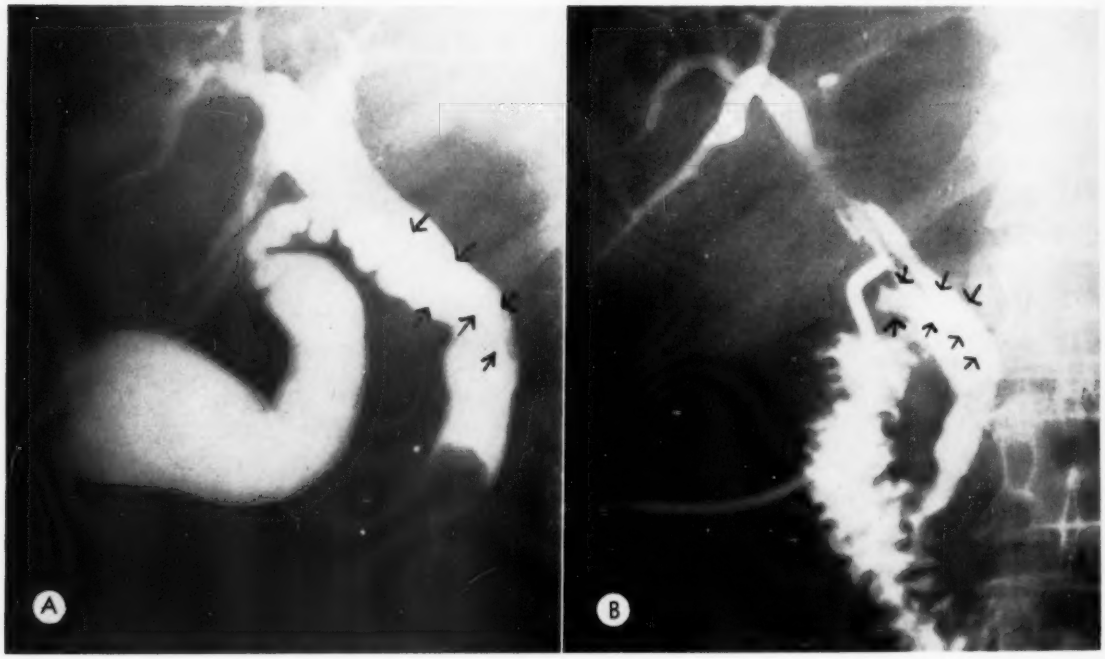

FIG. 3. (*A*) Roentgen examination of the biliary tree through a cholecystostomy. The intramural segment of the cystic duct can be seen within the projection of the common duct (arrows). There is dilatation of the biliary tree due to obstruction by the large gallstone visualized in the distal common duct. (*B*) Postcholecystectomy T-tube cholangiogram showing the intramural remnant of the cystic duct (arrows). Note its location at the medial aspect of the common bile duct.

this anomaly prior to intervening on the biliary system, especially if the contrast medium is introduced through a cholecystostomy (Fig. 3*A*). This procedure has been recommended by Hicken and coworkers[6] in order to perform an adequate cholecystectomy. Anatomic variations of the cystic duct can thus be visualized, ensuring its complete removal, if possible. Nevertheless, even after the identification of an intramural segment, its total removal may not always be accomplished by surgery. This is due to the close binding of the intramural portion of the cystic duct with the common duct.

Further evaluation of this seldom recognized condition is necessary and may be achieved in the future as newer contrast media are used for oral and intravenous studies of the biliary tree.

SUMMARY

An infrequently recognized variation of the cystic duct has been described. It consists of a short segment of the distal cystic duct which is intimately bound to the medial aspect of the common bile duct before opening into it. After cholecystectomy this segment is usually left behind and may cause recurrence or persistence of symptoms. The remnant can be identified by cholangiographic studies when the radiologist is familiar with such an anomaly of the extrahepatic biliary tree.

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CHOLEDOCHAL CYST*

REPORT OF A CASE WITH CONTRAST VISUALIZATION

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CHOLEDOCHAL cyst, a congenital localized dilatation of the common bile duct of unknown etiology, is relatively rare. Judd and Green[2] in 17,311 biliary tract operations from 1907 to 1926 encountered only 1 case of true common duct cyst. However, this pathologic condition has been known since the 18th century. According to Lavenson,[3] the first case of choledochal cyst was described by Vater in 1723.

The majority of the authors considers this condition to be congenital and Heiliger[1] reported a case in a stillborn fetus at term. It is postulated by some that the primary cause of the disease is a congenital anatomic weakness of the wall of the duct. Others feel that stenosing valve-like duplication of the wall or angulation in the distal portion of the duct plays a primary role in causing cystic dilatation. The combination of both seems most probable. Reported cysts vary in size from 1 inch to 12 inches in diameter, some occupying the entire abdomen. The distal portion of the common duct in the majority of cases is not involved. Roentgenographically, choledochal cysts have been demonstrated on plain roentgenograms as ill-defined masses in the right upper abdomen not well separated from the liver shadow. Aspiration of the cyst's contents and injection of contrast material as well as pneumoperitoneum have been done for diagnosis. Large cysts may show pressure defects on the duodenum and displacement of adjacent organs.

In 1957, Rheinlander and Bowens[6] described a case of congenital absence of the gallbladder associated with cystic dilatation of the common bile duct in which the operative cholangiogram permitted the visualization of the malformation with no contrast material passing into the duodenum. Exploration revealed a 2 cm. calculus in the left hepatic duct. Probing was unsuccessful in establishing a communication between the cyst and duodenum.

In most reported cases, oral cholecystography has failed to outline the dilated duct and rarely has the gallbladder been visualized. In 1935, Wright[8] reported a case with good visualization of the gallbladder which was displaced and deformed by a huge choledochal cyst. The cyst was not opacified by the contrast material. In 1957, Moseley[5] described a case of choledochal cyst visualized by oral cholecystography using telepaque as a contrast medium. In this case, both the gallbladder and the common duct cyst were well demonstrated. A gastrointestinal series following cholecystography showed the duodenum to be displaced, anteriorly and inferiorly, by the opacified cyst. These findings were confirmed by exploratory laparotomy. From a review of the literature, this is apparently the first report of roentgenographic demonstration of a choledochal cyst by oral cholecystography. In 1958, Liebner[4] reported the opacification of a choledochal cyst and gallbladder in an eight and one-half year old girl by both oral cholecystography and intravenous choledochography. Surgical exploration confirmed the roentgen findings.

Because of the rarity of contrast visualization of choledochal cysts, the following case seemed worthy of reporting.

REPORT OF A CASE

The patient, a twenty year old Negro female, was admitted to the Buffalo General Hospital on October 4, 1959 with a chief complaint of

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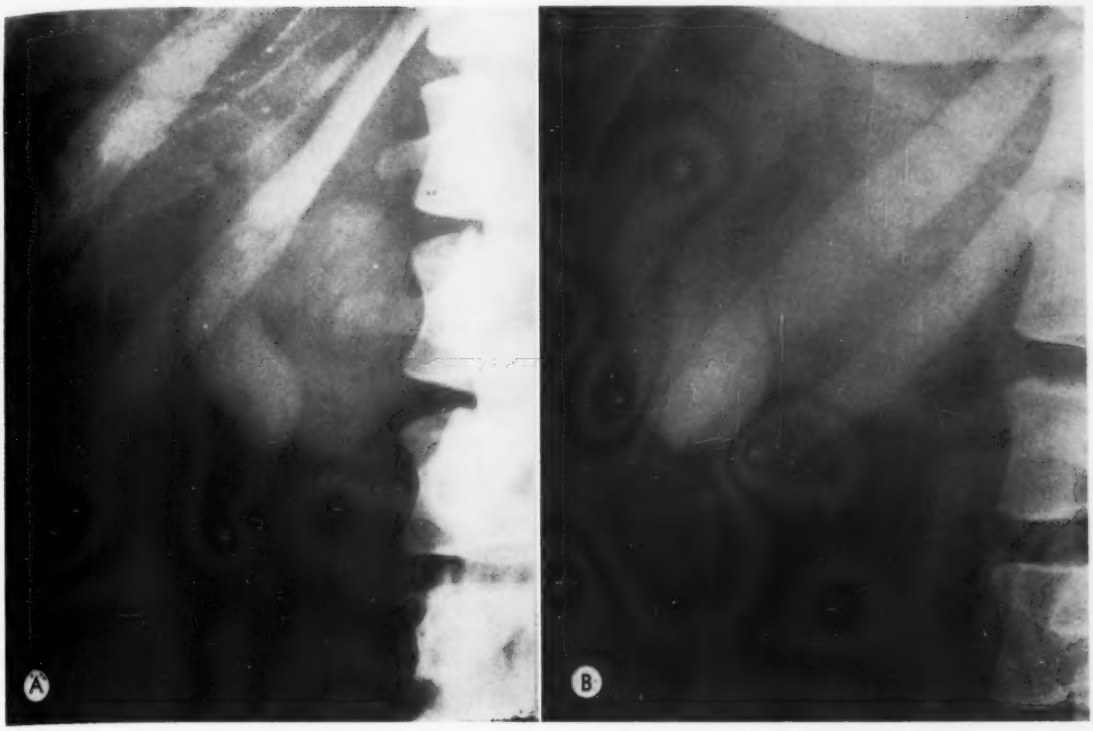

Fig. 1. Oral cholecystograms in (*A*) prone and (*B*) oblique projections.

right upper abdominal pain of four hours' duration associated with nausea and vomiting. Since the age of thirteen she had had intermittent attacks of abdominal pain classified by her as "stomach trouble." At the onset, when she was thirteen years of age, she underwent an appendectomy for these complaints. About two years prior to the present admission she had developed repeated attacks of right upper abdominal pain for which she was seen in the outpatient clinic. There had been two admissions to the hospital for pregnancy, one in March, 1958, and the second in September, 1959. Also she had been readmitted to the hospital after her first delivery because of severe right upper abdominal pain. She had multiple roentgenographic examinations which will be discussed later.

Physical examination on admission was not remarkable except for marked right upper abdominal tenderness. There was no spasm or rebound and no masses were palpated. All laboratory studies were negative. There was no evidence of clinical or chemical jaundice.

Roentgen Findings. Oral cholecystograms (Fig. 1, *A* and *B*) taken in March, 1958, showed a well-visualized gallbladder shadow. A second cyst-like structure lying medial and posterior to the gallbladder was opacified to about the same density. The interpretation at that time was a probable "double gallbladder." Upper gastrointestinal studies including the small bowel were normal. There was no evidence of any deformities or displacement by extrinsic masses. An oral cholecystogram taken in September, 1959 revealed findings identical with those of March, 1958.

Intravenous cholangiography was done when the patient entered the hospital in October, 1959. Cholangiograms up to three hours after the injection of cholegrafin showed the hepatic ducts and the proximal 3 cm. of a markedly dilated common duct. Cholangiograms taken at twenty-four hours are shown in Figures 2, *A* and *B*, and 3. Figure 4 is a composite diagrammatic sketch of the roentgen findings. It seemed quite obvious at this time that the large opacified cyst-like structure was a choledochal cyst.

Surgical Findings. Operation on October 13, 1959 disclosed a normal gallbladder with a normal cystic duct entering a dilated right hepatic duct which appeared to end blindly, and marked cyst formation. This cyst meas-

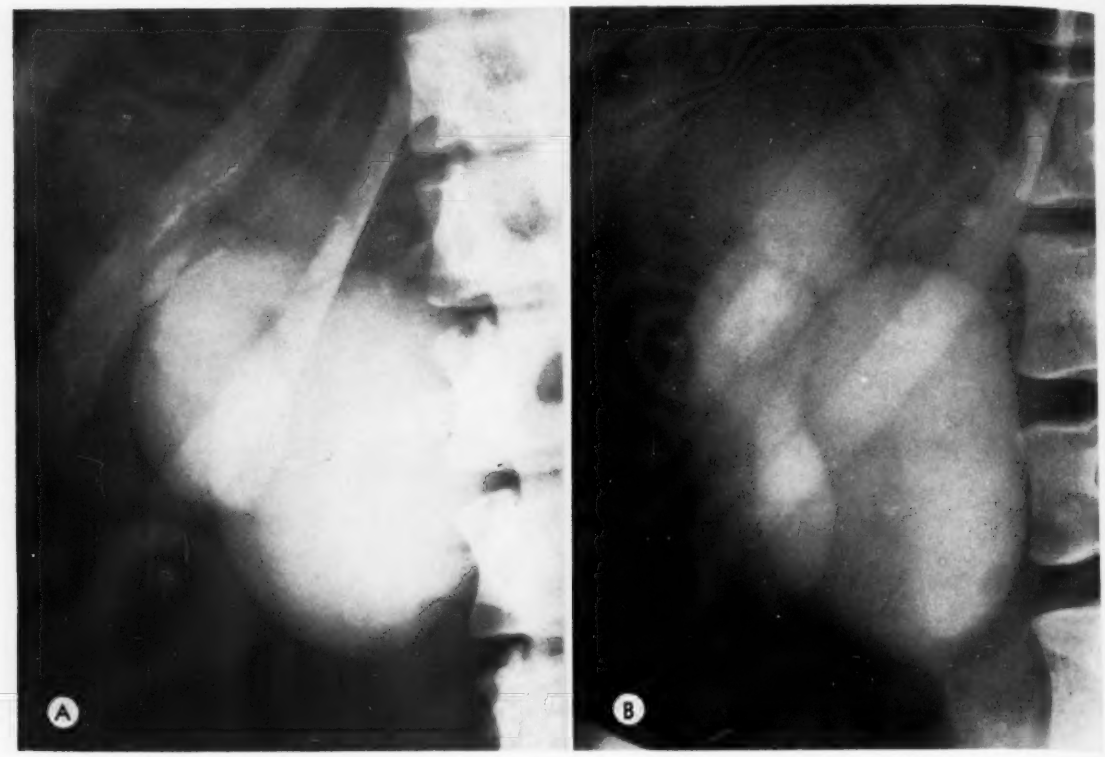

FIG. 2. Intravenous cholangiograms at twenty-four hours in (*A*) prone and (*B*) oblique projections.

ured approximately 5 inches in diameter and 300 cc. of bile was aspirated. An attempt to probe the duodenum through the cyst was unsuccessful. A normal-sized left hepatic duct was found. This had no communication with the cyst but passed downward toward the duodenum. Probing of this duct did not reveal a duodenal communication; however, it was the surgeon's opinion that such an opening did exist. (Figure 5 is a schematic drawing of the surgical findings.) An anastomosis was made between the cyst and a jejunal loop 10 inches beyond the first anastomosis. Biopsy of the cyst wall was histologically compatible with bile duct structure.

The patient's postoperative course was without complication and she was discharged to outpatient care on her tenth postoperative day. She has been relatively symptom-free since

FIG. 3. An intravenous cholangiogram in a far oblique projection shows the relation of the dilated right hepatic duct to the cystic dilatations below.

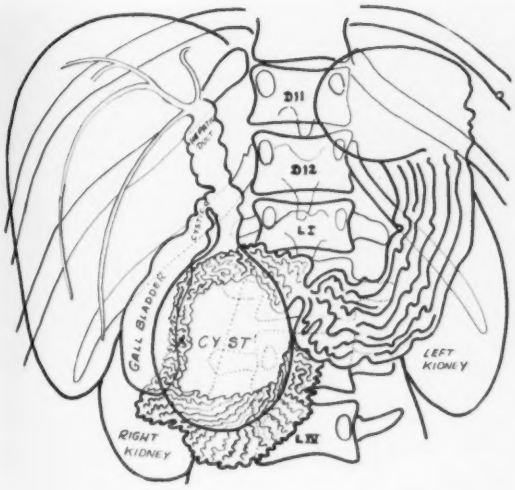

FIG. 4. Composite diagrammatic sketch of roentgen findings.

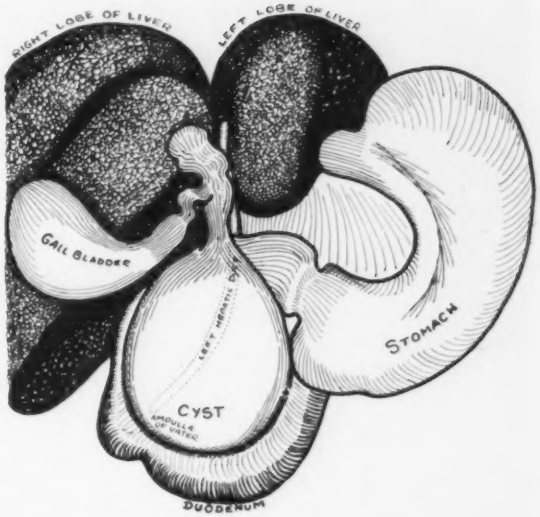

FIG. 5. Schematic drawing of the surgical findings.

surgery. She was last seen for follow-up in January, 1961.

DISCUSSION

The classic clinical triad of pain, tumor and jaundice is not always present with choledochal cysts. From a review of the literature, it is clear that frequently one component is absent and, in many instances, only abdominal pain is present. This was true in our case. At surgery frequently there is difficulty in identification and exploration of the distal bile duct. In our case it seems logical to assume that the cyst was arising from the right hepatic duct and, because of the absence of jaundice, that the left hepatic duct entered the duodenum.

Oral cholecystography is, in most instances, inadequate for demonstration of choledochal cysts. Probably, with the increased use of intravenously injected contrast media more cysts will be recognized and accurately diagnosed. Delayed filming may be of considerable importance for complete understanding of the structural changes. This was extremely helpful in the case reported here.

CONCLUSION

A case of choledochal cyst is reported which was opacified both by orally and intravenously administered contrast material. The superiority of the results using intravenous injection is stressed and it is pointed out that delayed filming may be highly important.

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THE COLLAGEN DISEASES: A DEMONSTRATION OF THE NONSPECIFICITY OF THEIR EXTRA-PULMONARY MANIFESTATIONS*

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THE broad spectrum of clinical and roentgen manifestations of the collagen diseases has been amply described.[1–6,8,9] Although the evidences of these diseases (disseminated lupus erythematosus, polyarteritis nodosa, dermatomyositis, scleroderma, rheumatic fever, and rheumatoid arthritis) are protean, each may present certain distinguishing features. However, they are often difficult to differentiate, particularly on the basis of the roentgen appearance of pulmonary involvement. We have felt, as have others, that the extrapulmonary roentgen findings in these diseases have more specificity, but more recent experience has suggested that these roentgen findings may be equally nonspecific and at times misleading.

This report is presented to illustrate these inconsistencies and to point out the broad overlap of the roentgen findings in these diseases.

REPORT OF CASES

CASE I. L.G. This thirty-four year old white woman was admitted June 3, 1958 with complaints of generalized weakness and gangrene of the right index finger. She had been hospitalized elsewhere in 1944 with a diagnosis of hemolytic anemia and had undergone splenectomy. She was readmitted to the same hospital in 1956, at which time a diagnosis of lupus erythematosus was made and confirmed by positive L.E. tests.

On her present admission, roentgen examination of the upper gastrointestinal tract showed atony and dilatation of the esophagus and small intestine (Fig. 1, *A* and *B*). Roentgen examination of the hands and wrists showed only osteoporosis. However, roentgenograms of the right forearm showed calcifications in the muscles of the proximal portion (Fig. 1, *C* and *D*).

The presence of soft tissue calcifications and the changes in the gastrointestinal tract were considered confirmatory of collagen disease and suggested scleroderma. Examination of the chest showed cardiac enlargement but no infiltration of the pulmonary parenchyma.

The patient expired shortly after admission. Autopsy confirmed the clinical impression of disseminated lupus erythematosus.

Comment: This patient with disseminated lupus erythematosus showed changes in the gastrointestinal tract and soft tissue calcifications, both of which are seen comcomly in scleroderma.

CASE II. B.J. This twenty-nine year old white woman was admitted on June 25, 1959 with complaints of weakness and fever of two months' duration. She also described intermittent dysphagia and joint pains. In May, 1959 she had been hospitalized elsewhere for evaluation and positive L.E. tests were found.

Roentgen examination of the upper gastrointestinal tract showed atony and dilatation of the esophagus (Fig. 2*A*). At fluoroscopy, the esophageal peristalsis was seen to be impaired and the distal end of the esophagus was patulous. The duodenal loop was dilated and also showed impaired peristalsis (Fig. 2*B*).

The patient's symptoms were fairly well controlled with prednisone and chloroquine but her condition gradually deteriorated and she expired approximately one year after her original admission. Autopsy showed disseminated lupus erythematosus with generalized vascular involvement.

Comment: This patient with disseminated lupus erythematosus showed changes in the gastrointestinal tract which mimicked those of scleroderma.

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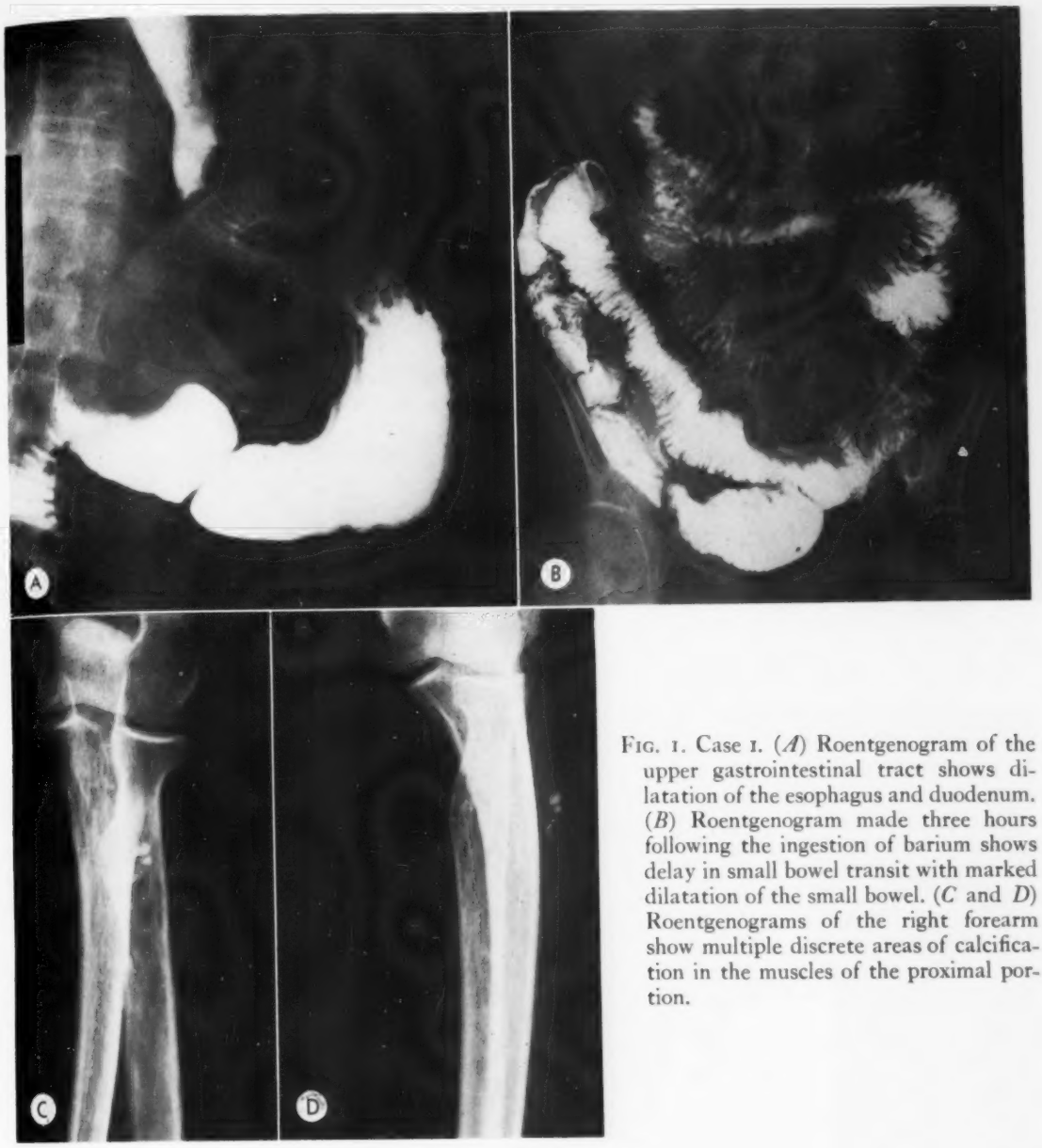

Fig. 1. Case 1. (*A*) Roentgenogram of the upper gastrointestinal tract shows dilatation of the esophagus and duodenum. (*B*) Roentgenogram made three hours following the ingestion of barium shows delay in small bowel transit with marked dilatation of the small bowel. (*C* and *D*) Roentgenograms of the right forearm show multiple discrete areas of calcification in the muscles of the proximal portion.

Case III. J.W.B. This sixty year old man was admitted on May 15, 1958 with the complaint of ulceration of the skin of his legs. During the previous thirteen years, he had noted thickening of the skin, starting on the left hand and extending to involve the skin of the arms, chest, abdomen, legs, and feet. Four years after the onset of disease, the skin of the hands and legs broke down and became ulcerated.

Physical examination showed ulcerations of the skin of both feet and ankles with old scars extending to the knees of both legs. There was no evidence of venous insufficiency.

Biopsy of the skin revealed scleroderma and cutaneous calcinosis.

Roentgen examination of the chest disclosed extensive linear infiltration throughout the parenchyma of both lungs. Examination of the lower extremities showed extensive calcification of both the skin and muscle (Fig. 3, *A* and *B*). The roentgen impression was dermatomyositis.

Comment: This patient with scleroderma

demonstrated extreme soft tissue calcification of the type usually seen in dermatomyositis.

CASE IV. T.S. This thirty-nine year old woman was admitted on April 2, 1958 complaining of arthritis of nine years' duration and chest pain of one and one-half years' duration. The arthritis was associated with swollen and painful joints of both hands, elbows, knees, and feet. The swelling and deformity were evident on examination.

Roentgen examination of the chest showed evidence of widespread infiltration of the pulmonary parenchyma. Examination of the hands and wrists revealed marked narrowing of all joint spaces and, in addition, there was evidence of destructive change in the lateral aspect of the head of the proximal phalanx of the third finger of the left hand (Fig. 4, *A* and *B*). Because of this destructive change, the roentgen impression was that of rheumatoid arthritis.

L. E. tests were positive on six different occasions.

Symptomatic relief was obtained with steroid therapy, but the disease followed a progressive course and the patient expired on March 3, 1959. Autopsy showed disseminated lupus erythematosus.

Comment: This patient with disseminated lupus erythematosus showed changes in the hands which closely resembled those of rheumatoid arthritis progressing to the stage of frank bone destruction.

CASE V. C.M. This sixty year old woman was admitted on February 17, 1959 with complaints of generalized arthralgia and muscle weakness of sixteen months' duration. These symptoms had progressed until the patient had become bedridden. Two months prior to admission she noted dysphagia.

Physical examination showed marked muscular weakness, the muscles being flabby and atrophic.

Roentgen examination disclosed atony of the pharynx and esophagus with absence of peristalsis, suggesting scleroderma (Fig. 5, *A* and *B*).

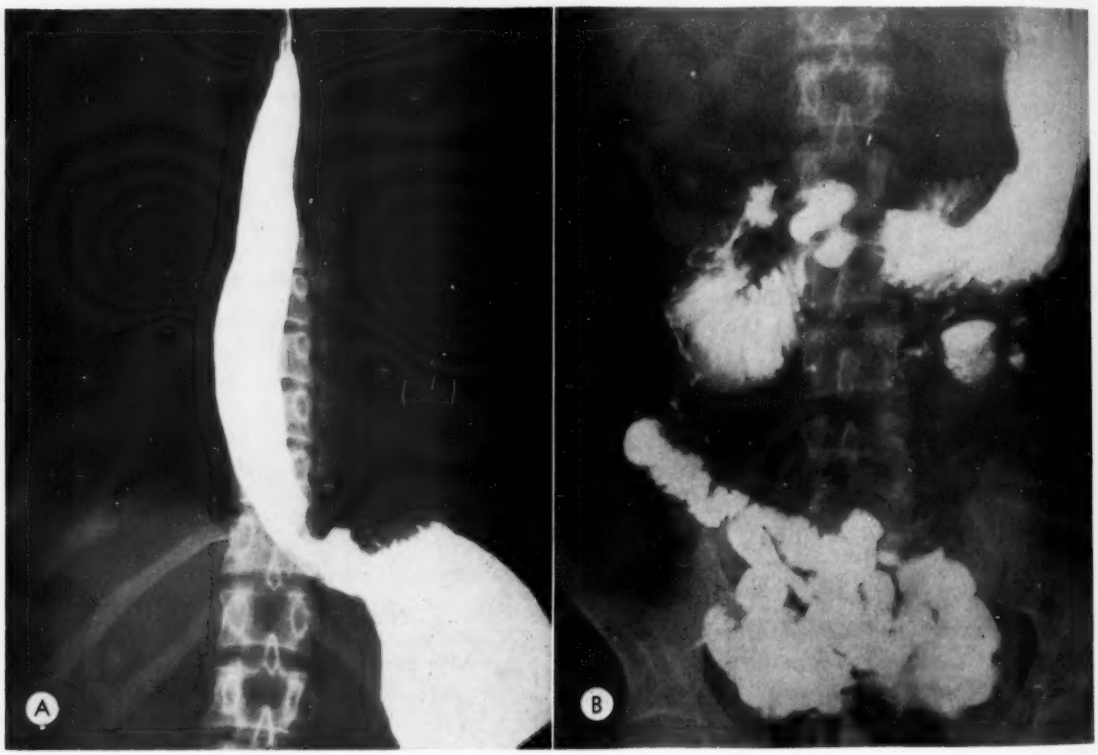

FIG. 2. CASE II. (*A*) Roentgenogram of the esophagus shows dilatation and atony. (*B*) Roentgenogram of the upper gastrointestinal tract shows dilatation of the duodenal loop.

Fig. 3. Case III. (*A* and *B*) Roentgenograms of both lower extremities show extensive calcification of skin and muscle and irregularity of the soft tissues indicating areas of ulceration.

Electromyography was performed with a reading consistent with a myopathy. The muscle biopsy was reported as dermatomyositis.

Steroid therapy resulted in definite improvement of muscle strength, and relief of dysphagia. The patient did not return for follow-up visits, but a report from her family physician stated that she died in May, 1959 of an exsanguinating gastrointestinal hemorrhage.

Comment: This patient with dermatomyositis showed pharyngeal and esophageal changes considered typical of scleroderma.

DISCUSSION

Each of the patients reported here exhibited one or more roentgen findings which are considered more typical of a form of collagen disease other than the one which was actually present.

Cases I and II with unequivocal lupus erythematosus revealed roentgen findings in the gastrointestinal tract and soft tissues which are identical to those described in scleroderma,[4] and this diagnosis was therefore suggested. In scleroderma the esophagus is widened and atonic, with diminished peristalsis, and regurgitation from the stomach occurs. The small bowel is dilated, with diminished peristalsis. Delay in transit is observed. The heart may be enlarged. Also, in scleroderma soft tissue calcification occurs, most often in the hands, but it may be present elsewhere, particularly in areas subjected to pressure, as in the forearms of our Case I. The similarity of these changes to those seen in our Cases I and II is striking.

It is true that the gastrointestinal tract

Fig. 4. Case IV. (*A* and *B*) Roentgenograms of both hands show osteoporosis, narrowing of joint spaces, and early destructive change in the proximal phalanx of the third finger of the left hand.

of patients with lupus erythematosus may show lack of tone, dilatation, and poor peristalsis, but usually these changes are much less severe than those seen in scleroderma and in our Case I.[3,6]

Case III, a patient with proved scleroderma, demonstrated extensive cutaneous and muscular calcification, which is more usually related to dermatomyositis than to scleroderma.[7,9] The calcification in this patient was not discrete in periarticular or pressure areas, as commonly is seen in scleroderma.[4] For this reason it resembled more the amorphous cutaneous and muscular calcification of dermatomyositis, the diagnosis which was suggested roentgenologically.

Case IV, a patient with proved disseminated lupus erythematosus, showed evidence of an inflammatory arthritis with osteoporosis and narrowing of the joint spaces of the hands. These osseous findings may be seen in lupus erythematosus, but, unlike rheumatoid arthritis, they do not as a rule progress to the phase of bone destruction.[2,9] The evidence of destructive change in one finger suggested the diagnosis of rheumatoid arthritis. Combinations of rheumatoid arthritis and systemic lupus erythematosus do exist,[2] but in this patient there was no other evidence to suggest such an association.

Case V, a patient with proved dermatomyositis, showed severe alterations in pharyngeal and esophageal motility. Although an aperistaltic esophagus has been described in dermatomyositis,[6] it is seen most consistently in scleroderma and usually suggests the latter diagnosis.

The data presented here are further evi-

dence for the concept that all of these diseases are systemic alterations of the connective tissue and that their pathogenesis may be similar. Since connective tissues can respond to injury in only a limited number of ways, it is not surprising that the clinical and roentgen manifestations of diseases of connective tissues may closely simulate each other and may, in fact, be indistinguishable.

These cases also illustrate the fallibility of over-reliance on any single roentgen change and the lack of specificity of these changes. A correct diagnosis requires the combined information derived from clinical, roentgen, and laboratory sources.

SUMMARY

Five cases of collagen disease (3 of systemic lupus erythematosus, 1 of scleroderma, and 1 of dermatomyositis) are reported. Each of these cases presented extrapulmonary roentgen findings which are, as a rule, more typically seen in some other form of this family of diseases. Clinically and roentgenologically, these diseases are poorly defined and undue reliance should not be placed on a single roentgen manifestation.

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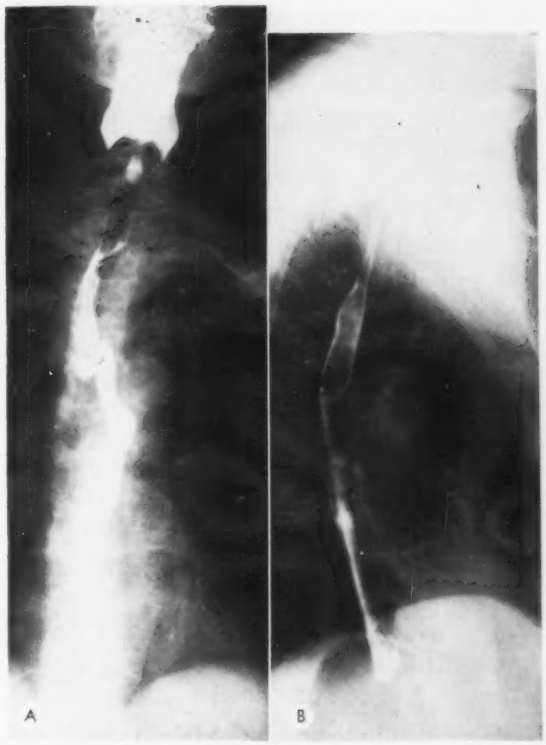

FIG. 5. Case v. (*A* and *B*) Roentgenograms of the esophagus show stasis of barium in the pharynx and esophagus and absence of peristalsis.

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THE ROENTGEN APPEARANCE OF MUTILATING PALMO-PLANTAR KERATOSIS*

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THE disease characterized by the formation of disabling hyperkeratoses of the skin of the palms and soles is best known as palmo-plantar keratosis or keratoma, but it is also called by the following synonyms: keratoderma, hyperkeratosis palmaris et plantaris,[1,2] tylosis palmarum et plantarum, ichthyosis palmaris et plantaris, symmetric keratodermia of the extremities,[4] and Méléda disease.[3] The multitude of synonyms is due in part to the difference in degree and extent of the disease process and to the other ectodermal dysplasias which may be associated with it.[7] The mutilating form of this disease is rare, however, and to our knowledge the case reported herein is the most extensive yet described. The mutilations of palmo-plantar keratosis offer characteristic roentgenographic changes from which the diagnosis can be made or strongly suspected.

HISTORICAL REVIEW

In 1826 Stulli described an endemic, congenital disease occurring in the inhabitants of the island of Méléda in southern Dalmatia which was manifested by hyperkeratosis involving chiefly the palms and soles.[4,8] Stulli's cases were initially thought to be endemic leprosy, but were later identified as the same palmar and plantar keratoma described by Thost in 1880 and Unna in 1883.[8] The hereditary and familial nature of the disease probably explains the high incidence on Méléda, where intermarriage occurred frequently in an isolated community.[4]

ETIOLOGY AND CLINICAL COURSE

Palmo-plantar keratosis is characterized by symmetric thickening of the horny layer of skin beginning on the palms and soles and occasionally extending over the dorsal surfaces, especially about the metacarpophalangeal joints.[4,6] It may be congenital or acquired. The acquired form, an uncommon variant, appears to be secondary to eczema, infection, psoriasis or toxic dermatoses (arsenic).[3] In the congenital form heredity is the important factor. Many authors have cited multiple cases occurring through several generations of a given family.[4] Turpin *et al.*,[7] for example, followed the development of the disease through six generations in 15 members of a family of 28 individuals. He suggested that the genetic character is monogenic, autosomal and dominant.

Congenital palmo-plantar keratosis is rarely manifest at birth and is generally not recognizable until the third or fourth month of life. Occasionally it may not appear before four or five years of age, even when of congenital origin. The hyperkeratosis begins as patches of epithelial thickening which grow and coalesce until they form thick, smooth, yellowish-white, translucent, hard plates over the palms and soles. The palms are eventually completely covered by these plates, while, as a rule, only the weight-bearing portions of the soles are involved. The horny plate becomes rigid and inelastic, predisposing to the formation of fissures.[3,4] Further, in both the congenital and acquired forms of keratoma, hyperhidrosis soon develops, resulting in maceration and additional fissuring of the lesions.[3–5,7–9] With maceration comes infection, and the hyperkeratoses lose their plate-like appearance and assume a dirty gray to brown color and a verrucous surface, and they may become odoriferous. Hyperkera-

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totic lesions may later appear on the dorsa of the hands, and on the knees, elbows, in the groin, or on any other area of the skin at which pressure due to weight bearing or clothing abrasion may occur.[8]

In certain severe cases, mutilation in the form of spontaneous amputation of digits may occur.[8,9] This seems due in part to the development of deep, annular, horny, constrictive bands which form about the digits and which seem to be peculiar to the mutilating form of palmo-plantar keratosis. Additional mutilation may occur as the result of fissuring, infection and consequent trophic complications due to scarring and pressure upon nerves and vessels.

The histopathology is nonspecific. The skin layers are uniformly thickened with excessive cornification and may be otherwise normal.[4] The lesion may be confused pathologically with psoriasis. When fissures have developed, infllammatory changes are present because of secondary infection.

In the usual form of palmo-plantar keratosis, roentgenographic findings do not ordinarily contribute to the diagnosis, but, when the mutilating form is present, roentgen manifestations characteristic of this entity will appear and they are described below.

REPORT OF A CASE

R.R., a Negro male, was born September 20, 1941, the third of four children. Although apparently normal at birth, at two months of age he developed a persistent, rough, erythematous, scaly rash over the inguinal area and buttocks. At eighteen months, patches of dry, scaly, thickened skin appeared on the soles and similar lesions on the palms three months later. These hyperkeratoses rapidly spread to involve the entire palmar, and much of the plantar, surfaces. The disease process continued and, at the age o e years, the skin over the palms, soles, inguinal and perineal regions was scaly and thickened with layers of dead skin. The extremely thick keratoses of the palms and soles caused swelling and spreading of the digits. At that time, the left hand showed amputation of the distal phalanx of the fourth finger and a wet granulating stump of the distal phalanx of the fifth finger. In addition, the skin over the knees and elbows showed keratotic plaques. There was kyperkeratotic skin about the corners of the mouth, the ears, the penis and scrotum.

Roentgenograms of the extremities made at the age of five are said to have shown demineralization of the metacarpals and phalanges with resorption of the tufts of all the distal phalanges. There were destructive lesions involving several of the other phalanges, and, of course, the amputations were apparent.

At the age of eight the distribution of the lesions was the same but the hyperkeratoses had become larger, forming dense, fissured, woody masses which extended deep into the palms. The hands were claw-like and functionless. The fingers and thumbs showed progressive atrophy with proliferation of keratin tissue about the nails, and on the basis of this latter change an erroneous diagnosis of pachyonychia congenita was made. The distal portions of the fingers were atrophic and about the size of an infant's digits. The feet showed the same findings as the hands with slightly less atrophy of the digits. Several more distal phalanges of hands and feet were absent.

Pathologic examination of a biopsy specimen from the sole of the left foot showed marked hyperkeratosis with piling up of excessive keratohyalin material on the epithelial surface. Epithelial hyperplasia was seen with widening of the stratum granulosum and elongation of the rete pegs. Diffuse chronic inflammatory infiltration was present.

When he was eight years old the patient had prolonged hospitalization for surgical procedures on the feet which included amputation of the remaining toes and resection of the plantar surfaces. Pedicle grafts were brought to the under surfaces of the feet from the thighs. Due to infection, the right foot had to be grafted twice. Then, as happened so many times throughout his course, the patient was lost to follow-up for a lengthy period.

At the age of fourteen the patient was admitted to the hospital twice for staphylococcal septicemia arising from the infected extremities. At that time the skin grafts on the plantar surfaces were intact, and, of course, the toes had been previously amputated. All fingers were now missing from the hands as a result of autoamputation, and, due to the development of hyperhidrosis, macerated, fissured skin covered palmar and dorsal surfaces.

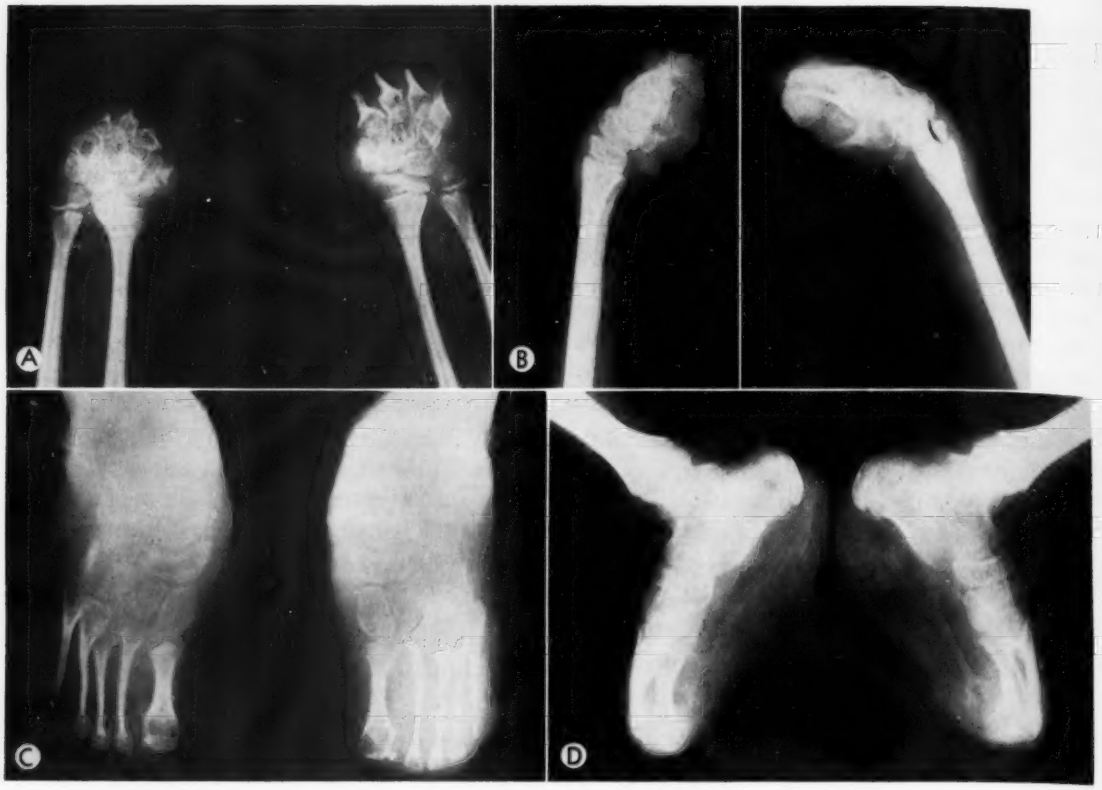

FIG. 1. (*A*) Posteroanterior roentgenograms of the wrists and hands, made at the age of fourteen, show preservation of mineralization of bones, but there is amputation of bones and soft tissues at the level of the metacarpal bases. There is some thinning of the distal shafts of the radii and ulnae. The remaining soft tissue of the hands appears somewhat irregular. (*B*) Lateral roentgenograms of the hands and wrists show thick, featureless soft tissue over the palms. Surgical dressings are in place. (*C*) Anteroposterior roentgenograms of the feet, made at the age of fourteen, show amputations of all toes at the level of the metatarsal heads. Mineralization is, in general, good, although there is trophic thinning of the shafts of all metatarsal bones. (*D*) Lateral roentgenograms of both feet show thickening of the soles due to replacement of the keratomatous plantar tissue by fatty skin grafts. All prints were made from microfilm reproductions.

Roentgenograms of the extremities made at the age of fourteen (Fig. 1, *A–D*) show slight demineralization of bone. Autoamputation, probably a result of cornified bands, the pressure exerted by abnormal tissue masses, and inflammatory scarring, has extended to the level of the metacarpal bases in the hands (Fig. 1, *A* and *B*) and to the metatarsal heads in the feet (Fig. 1, *C* and *D*). Figure 1*D* shows the thick, fatty skin grafts applied over the soles of the feet six years earlier.

The patient returned recently, now nineteen years old, and was seen by us for the first time. There was abundant evidence that the destructive process had continued. The hands and feet were now macerated stumps partially covered with warty, nodular keratotic plaques (Fig. 2, *A* and *B*). The skin graft on the left foot was largely replaced by verrucous, macerated keratoma tissue, but the right skin graft was intact with keratomatous change about its margins (Fig. 2, *C* and *D*). Hyperkeratotic plaques were present on the skin about the knees (Fig. 2*C*), elbows and sacrum. There was hyperkeratosis to a lesser degree under the arms, about the groin and about the ears. Review of these findings, and of pertinent dermatologic literature, led us to the diagnosis of mutilating palmo-plantar keratosis.

Roentgenograms made at the present time (Fig. 3, *A*, *B* and *C*) show that there has been progressive loss of bone during the past five years (compare with Figure 1, *A–D*). All metacarpals have now been lost from the hands (Fig. 3*C*) and there is fusion of the remaining carpal elements, possibly the end result of

superimposed bone infection. Note that mineralization is rather well preserved. There is thinning of the distal aspects of the shafts of the left radius and ulna, and periosteal reaction, perhaps the product of infection, is seen along the distal medial ulnar shaft. Especially noteworthy is the appearance of the soft tissues on the roentgenogram, for one can see the nodular, warty appearance characteristic of infected, macerated keratoma tissue. These findings are even more striking on the roentgenograms of the left foot. There has been further autoamputation of all of the metatarsals and some of the tarsal bones in the left foot (Fig. 3*A*) and of the distal second, third and fourth metatarsals in the right foot (Fig. 3*B*). Note the trophic thinning of the shafts of the first and fifth right metatarsals (Fig. 3*B*).

The patient himself is mentally alert and intelligent, and he can use his upper extremity stumps to feed himself and to write. Future management will include removal of the stumps,

Fig. 2. (*A*) Photograph of the dorsal surfaces of the arms, made at the age of nineteen. Note destruction of the hands and replacement by nodular, fissured, macerated tissue. (*B*) Volar surfaces of the wrists and hands showing warty, macerated keratomatous tissue.

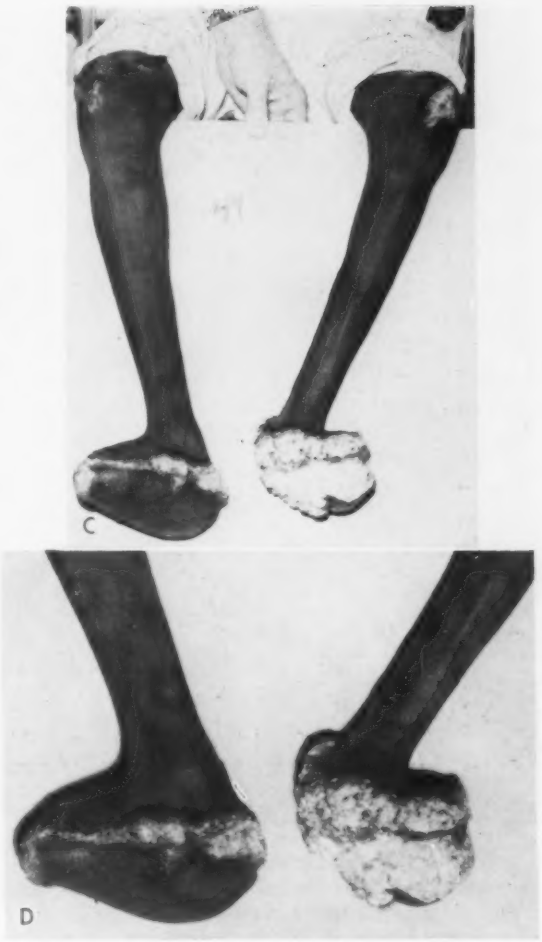

Fig. 2. (*C*) Photograph of the legs and feet, made at the age of nineteen, shows replacement of the left plantar graft by keratomatous tissue which has appeared along the margins of the still-preserved graft on the sole of the right foot. Note hyperkeratotic lesions over the knees. (*D*) Close-up of the feet.

which are unsightly and a source of infection, and the development of prosthetic devices which will fit in such a way as to exert minimum pressure on the skin and so avoid the development of new areas of kyperkeratosis. Thorough questioning of the patient's family yields no history of skin lesions of any sort.

DISCUSSION

Palmo-plantar keratosis is an ectodermal defect, commonly, perhaps always, congenital in origin, manifested by a tendency toward excessive cornification of the skin of the palms and soles as well as at other pres-

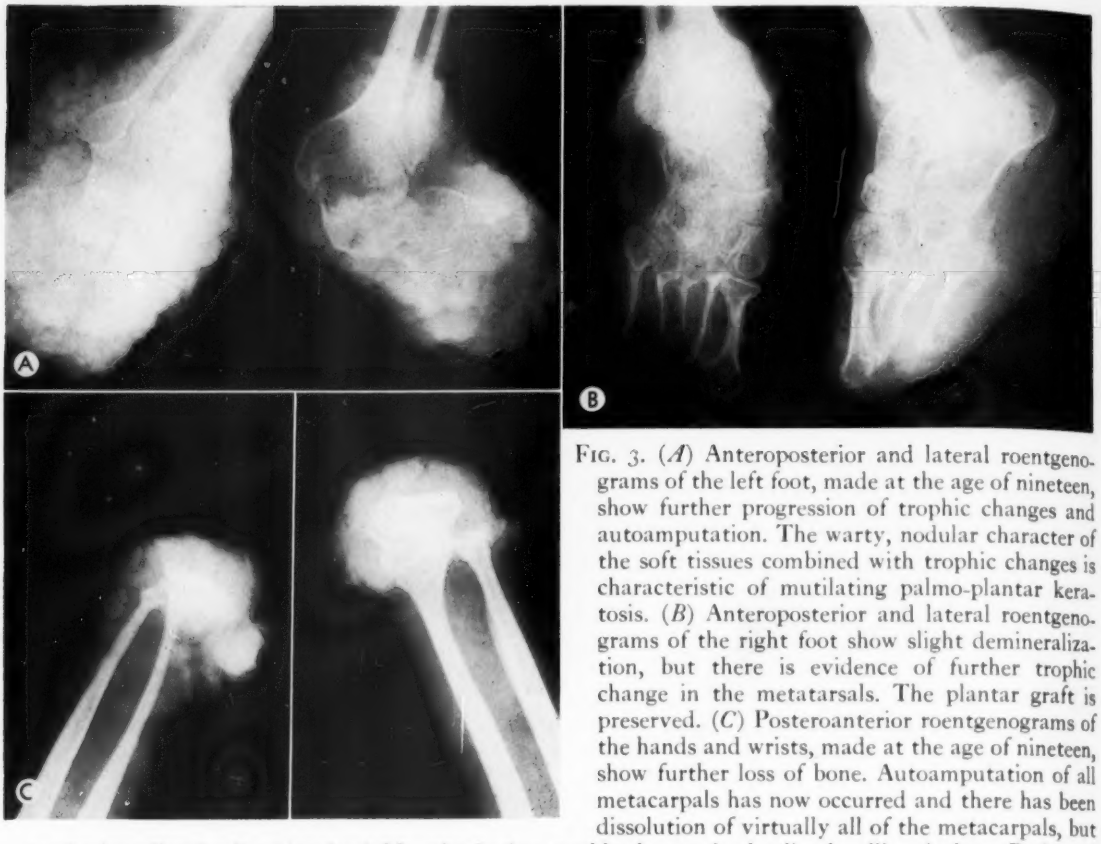

FIG. 3. (*A*) Anteroposterior and lateral roentgenograms of the left foot, made at the age of nineteen, show further progression of trophic changes and autoamputation. The warty, nodular character of the soft tissues combined with trophic changes is characteristic of mutilating palmo-plantar keratosis. (*B*) Anteroposterior and lateral roentgenograms of the right foot show slight demineralization, but there is evidence of further trophic change in the metatarsals. The plantar graft is preserved. (*C*) Posteroanterior roentgenograms of the hands and wrists, made at the age of nineteen, show further loss of bone. Autoamputation of all metacarpals has now occurred and there has been dissolution of virtually all of the metacarpals, but good mineralization is maintained. Note beginning trophic changes in the distal radii and ulnae. Periosteal reaction along the medial aspect of the distal left ulna may be the result of osteomyelitis from the always-prevalent soft tissue infections. Soft tissues show the characteristic irregular, lumpy appearance of far-advanced palmo-plantar keratosis.

sure points about the body. The most morbid form of this disease, apparently a rare complication, will progress to mutilation in the form of autoamputation of phalanges and soft tissues of the digits, later extending to metacarpal and metatarsal regions. Autoamputation of digits may be caused by circumferential cornified bands with embarrassment of blood supply and innervation, but the destructive changes in the palms and soles, and probably to some extent in the digits as well, seem also to be the result of pressure on bone, nerves and vessels by keratomatous masses, and possibly of inflammatory scarring produced by repeated infections that occur in the deep clefts of the macerated, verrucous keratoma tissue. In the case reported above, yet another type of bone change was present. It is illustrated by the fusion of carpal elements seen in Figure 3*C* and is probably the end result of healed osteomyelitis and infectious arthritis transmitted from infections constantly present in the abnormal overlying soft tissue.

There is a lengthy list of disease entities which can cause terminal phalanx or digit resorption, including collagen diseases such as scleroderma and rheumatoid arthritis, and such processes as Raynaud's disease, thromboangiitis obliterans, syringomyelia, frostbite syndrome and ergot poisoning. Seldom does bone atrophy in these conditions extend to proximal phalanges or beyond. Digital autoamputation can occur in the fifth, and occasionally the fourth, toe in ainhum, but probably the disease most seriously to be considered in the differential

diagnosis of the findings described in our case above is leprosy. In the neural form of leprosy, progressive distal atrophy and autoamputation can occur, as can soft tissue ulceration and secondary osteomyelitis. A useful objective roentgen differential diagnostic point is the identification of the nodular, warty keratoma tissue, well illustrated in Figure 3, *A* and *C*, which is the product of fissuring of the macerated horny plates produced by the dystrophic epithelium in palmo-plantar keratosis.

As a matter of interest, it can be said that treatment of palmo-plantar keratosis remains supportive, with the application of softening unguents to prevent cracking of the horny epithelial plates present initially, the administration of antibiotics for deep infections which may occur, and surgical relief of digital compression bands. In the case we have described this treatment could not be carried out, but whether it would have been of avail in preventing the mutilation which has occurred is not known.

SUMMARY AND CONCLUSIONS

1. A congenital familial disease, palmo-plantar keratosis, is initially manifested by dense thickening of the epithelium of palms and soles, and of the skin at pressure points about the body. Later, maceration may occur as the result of hyperhidrosis.

2. A morbid variant of this disease may produce mutilation in the form of autoamputations of digits, and finally of the entirety of the hands and feet.

3. The findings of trophic bone atrophy combined with regularly nodular, verrucous soft tissue are thought to afford a relatively absolute basis for roentgenologic identification of this entity.

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SOME ROENTGEN FEATURES OF PAGET'S DISEASE*

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PAGET'S disease, an entity of unknown etiology, is found in over 3.5 per cent of the population beyond the age of forty-five years.[5,19,20] The incidence increases until the age of seventy-five after which it levels off. The disease affects males more often than females[20] and shows a familial predisposition.[10,17] Most cases are detected initially as incidental findings on roentgenograms obtained for other purposes. Other patients may see a physician because of pain, fracture, deformity of an extremity, increased head size, or warmth of the overlying skin. Some cases are discovered by dentists when they find hypercementosis or ankylosis of a tooth, or deformity of the jaws. Deafness from involvement of the inner ear or signs of malignant degeneration of an involved bone may be the first indications of the disease.

The disease is localized to one or more bones, although, rarely, all, or almost all, of the bones may be affected.[1] According to Dickson *et al.*,[7] the pelvis is the most common site of the disease, with involvement of the femur, spine, skull, and tibia next in frequency, respectively (Table 1).

TABLE I

LOCATION OF OSTEITIS DEFORMANS IN 367 CASES*

Site of Involvement	Cases†	Combined Phase‡ (100 cases)	Sclerotic Phase‡ (100 cases)
Pelvis	243		
Iliac bones	—	107	100
Ischia	—	97	73
Pubic bones	—	96	73
Femur	171	68	40
Skull	153	—	—
Tibia	127	51	22
Sacrum	—	42	35
Lumbar spine	103	38	30
Dorsal spine	51	16	9
Clavicle	33	8	15
Humerus	20	9	1
Ribs	12	2	—
Scapula	7	1	—
Radius	7	1	—
Cervical spine	6	2	—
Fibula	6	2	—
Ulna	5	1	—
Os calcis	4	1	2
Patella	4	2	—
Talus	1	—	1

* Reprinted with permission of *Radiology*.[7]
Not including bones of the face and base of skull. More than one site of involvement in a number of cases.
† Cases of involvement of given site among total 367 cases.
‡ Classified roentgenographically. In these columns bilateral involvement is counted twice.

PATHOLOGY

Macroscopically, in advanced cases, the involved bone is thickened and presents a rough, uneven surface. Long bones are frequently deformed; compact bone is replaced by lamellated spongy bone of varying density, and the marrow cavity is wide or narrow. In cranial involvement, the bones of the vault of the skull are sometimes greatly thickened. The inner and outer plates and diploë may not be distinguishable, with the entire thickness consisting of spongiosa. If the base of the skull is involved, it sometimes bulges into the cranial cavity. The bones and periosteum are frequently vascular and the vessels leading to the bones are enlarged.[8,15]

Paget's disease is primarily one of bone destruction. Osteoclastic resorption of the trabeculae of the spongiosa or the walls of the haversian canals of the compacta is followed by connective tissue proliferation. Immature membranous bone is formed, which is destroyed and reconstituted repeatedly, producing the irregular, numerous cement lines and characteristic mosaic pattern described by Schmorl.[20]

* From the Department of Radiology, University of California, School of Medicine, San Francisco, California. This work was supported in part by Research Grant A-2589 GM, National Institutes of Health, U. S. Public Health Service.

Extensive involvement stimulates the periosteum to produce bone which is resorbed and converted to Paget's bone, resulting in expansion of the cortex.

The destructive process is self-limited and almost invariably is followed by osteogenesis, which is a manifestation of healing. Osteogenesis is stimulated by stress, such as weight bearing, and is inhibited by immobilization to a greater extent in Paget's disease than in normal bone.

The serum alkaline phosphatase, which represents the degree of osteoblastic activity, is elevated in cases with extensive involvement and active disease. It is least elevated in the healed or sclerotic phase. The sedimentation rate is also frequently elevated with active Paget's disease.[11]

PATHOLOGIC FEATURES OBSERVED ROENTGENOGRAPHICALLY

Many of the pathologic features of the disease can be observed roentgenographically, particularly if serial roentgenograms made over a period of years are available. One such process, that resulting in the deformity of bones present in advanced cases, can be observed readily in the sequence of events leading to lateral curvature of the femur or anterior curvature of the tibia. Weight bearing produces tension forces upon the lateral surface of the femur and compression forces on the medial surface.[9] The extremely active and weakened Paget's bone responds with periosteal apposition on the surface subjected to tensile forces, leading to a lateral expansion of the bone. This process may be followed by resorption of the opposite cortical surface proceeding at an almost equal rate and by compensating apposition and resorption of the endosteum, which maintains the medullary canal (Fig. 1). The curvature of the tibia is the result of bone apposition on the anterior surface followed by resorption of the posterior surface (Fig. 2 and 3). Here too, apposition is on the side of the bone subjected to tension, and resorption on the side where compression exists. This explanation also accounts for elongation of the bone. It is readily apparent in the curved tibia that the bone is longer than normal, as, despite the curvature, the upper and lower ends maintain their relationship with the normal fibula. A similar mechanism is assumed to be responsible for the curvature of other long bones (Fig. 4), the basilar invagination of the skull, and inward protrusion of the acetabula and adjacent structures in the pelvis (Fig. 5). The forces responsible may be due to muscle pull or gravity.

Fig. 1. Increased convexity of the left femur from bone apposition along the lateral surface. Slight resorption of medial subcortical surface has produced curvature. Bone deposited upon the medial endosteal surface, without corresponding resorption of the lateral endosteal surface, results in a narrow medullary canal.

The process of apposition and attrition of bone is frequently seen in migrating teeth and in the enlarged sella turcica caused by a pituitary tumor. It operates normally in the enlargement of the calvarium with growth of the brain.

FIG. 2. Bone apposition on the right tibia anteriorly without resorption of the posterior surface.

TABLE II

INCIDENCE OF PATHOLOGIC FRACTURE IN 367 CASES OF OSTEITIS DEFORMANS*

Location of Fracture	Patients
Thoracic portion of spinal column	24
Lumbar portion of spinal column	24
Right femur	8
Left femur	1
Right tibia	6
Left tibia	3
Right humerus	1
Left humerus	1
Separation of tibial tubercle	2
Ischium (only ununited fracture observed)	1
Basioccipital protrusion (extensive)†	6
Total number of pathologic fractures among 62 patients	77

* Reprinted with permission of *Radiology*.[7]

† The present author does not consider basioccipital protrusion to represent a fracture.

FRACTURES

The bones in Paget's disease, although sometimes appearing strong, are much more prone to fracture than normal bone when subjected to a bending force, particularly during the destructive phase.[2,9,11] The fractures may be complete, are usually transverse, or occasionally oblique, and most frequently occur in the spine or upper part of the femur and tibia (Table II). Collins[5] found 9 fractures in 70 patients, but only 1 in a routine survey of 650 consecutive patients, 24 of whom had Paget's disease. He estimated that the incidence must be less than that indicated in published reports and not more than 1 per cent of all cases.[5] The fractures heal readily, the

FIG. 3. Marked anterior bending deformity of the left tibia due to bone apposition anteriorly and resorption posteriorly. An almost equal apposition on the posterior endosteal surface and resorption of the anterior endosteal surface maintains the width of the medullary canal. Although curved, the ends of the tibia maintain an almost normal relationship with the fibula, indicating increased bone length.

callus converting to the same type of mosaic structure as the surrounding bone.

The fractures in the femur and tibia are more often incomplete, extending from the periosteum to the medulla in the shaft or for varying distances into the cancellous bone at the ends. These fractures occur along the lateral surface of the femur or the anterior surface of the tibia. They appear on roentgenograms as fissures lying perpendicularly to the long axis of the bone (Fig. 6*A*). Occasionally, if they have been present for several days or weeks, the margins of the defects are sclerotic. The incomplete fractures have been confused with the pseudofractures of osteomalacia but can usually be differentiated by their position (Fig. 17). Pseudofractures in osteomalacia involve the medial surface of the shaft or superior surface of the neck of the femur; they are associated with generalized bone disease and frequently are almost symmetric bilaterally.[14,25]

FIG. 4. Bending and elongation of the radius due to periosteal and endosteal apposition and resorption; also slight bone growth distally as the articular surface and styloid process of the radius extend beyond the usual relationship with the ulna.

FIG. 5. Bending inward of the medial wall of the right side of the pelvis due, primarily, to bone apposition. Some resorption of the acetabular surface, indicated by migration of the femoral head medially. The uniformly narrow articular cartilage of the right hip is presumably due to the resumption of endochondral ossification without corresponding cartilage proliferation. The early, primarily destructive stage of Paget's disease is demonstrated in the upper end of the left femur and in the left innominate bone. There are thickened bone bundles along the lines of stress.

Two factors are probably responsible for the location of the incomplete fractures: (1) When a sudden or heavy load is exerted upon the lower extremity, the tension or stretch exerted along the lateral aspect of the femur and the anterior surface of the tibia breaks the structurally weak Paget's bone. The forces along the opposite surfaces of these bones are those of compression and, therefore, do not produce fractures. (2) The fractures tend to occur in the region of bone resorption at the advancing margin of the lesion. The disease usually begins at the upper end of the femur or tibia and it is here, where the tensile forces are greatest, that the highest incidence of fractures is found. Occasionally the disease originates at the lower end, or at both ends simultaneously, and, regardless of the site of origin, as the disease spreads, additional fissure fractures occur at intervals, some-

FIG. 6. (*A*) Lateral roentgenogram of the tibia obtained after the patient experienced sudden pain. The lower 2 of 4 horizontal lines visible are horizontal fractures of the anterior cortex, but they extend obliquely downward and posteriorly to involve the posterior cortex. Soft tissue swelling was visible adjacent to the fractures. The upper two lines, covered by bone along their endosteal and periosteal surfaces, represent residual defects from old fractures. (*B*) Roentgenogram several months later. Prompt healing of posterior extensions of the fractures, probably because the fragments were approximated by compressive forces. The persistent anterior defects have joined the upper two defects as permanent scars.

times almost equidistant from each other (Fig. 7).

The fresh cortical fractures are associated with pain. When the anterior surface of the tibia is involved, a localized soft tissue swelling is visible on lateral roentgenograms. Shortly after the fracture is sustained, the periosteum forms bone over the fracture and for a variable distance surrounding it, leading to further thickening of the cortex. Endosteal bone also forms, limited to the immediate vicinity of the fracture and encroaching upon the medullary canal. Thus, the ends of the fractures are covered but the bony defect in the center of the cortex remains as a permanent scar. In some cases, a series of such scars attest to numerous previous traumata. The scars can be differentiated from recent fractures because they do not extend out to the periosteal surface of the cortex (Fig. 6, *A* and *B;* and 7). Although fissure fractures with complete healing were reported in the earlier literature, this was not true in our experience, possibly because our patients usually were treated on an ambulatory basis or with limited immobilization to avoid such complications as increased destruction of bone, hypercalcemia, and renal calculi. Perhaps continued weight bearing may prevent complete healing and lead to fibrous union. Where fractures extended completely across the shaft of a bone, the surface subjected to compression forces healed early, whereas defects persisted on the opposite surface (Fig. 6*B*).

CYST-LIKE DEFECTS

Radiolucent cyst-like defects of various size, surrounded by dense bone, may be seen in the spongy bone of the pelvis, the head and neck of the femur and other bones, and in deep layers of compact bone. These are collections of fat, resulting from necrosis and liquefaction of the proliferating fibrous tissue.[12]

CLASSIFICATION

The roentgen appearance may be classified into three descriptive forms: (1) sclerotic, (2) mixed, and (3) radiolucent. Multiple bone involvement may show similar or dissimilar lesions. Sometimes a single bone my exhibit more than one type of lesion.

Sclerotic Form. When present in cancellous bone, this consists of a uniform thickening of the trabeculae with a consequent

FIG. 8. Sclerotic Paget's disease of the left innominate bone.

FIG. 7. A series of cortical defects along the lateral surface of the femur, representing healed fractures. Bone covers their periosteal and endosteal surfaces. Characteristically, the periosteal new bone is smooth surfaced, whereas the endosteal surface is irregular because of bone repair limited to the immediate region of the fracture.

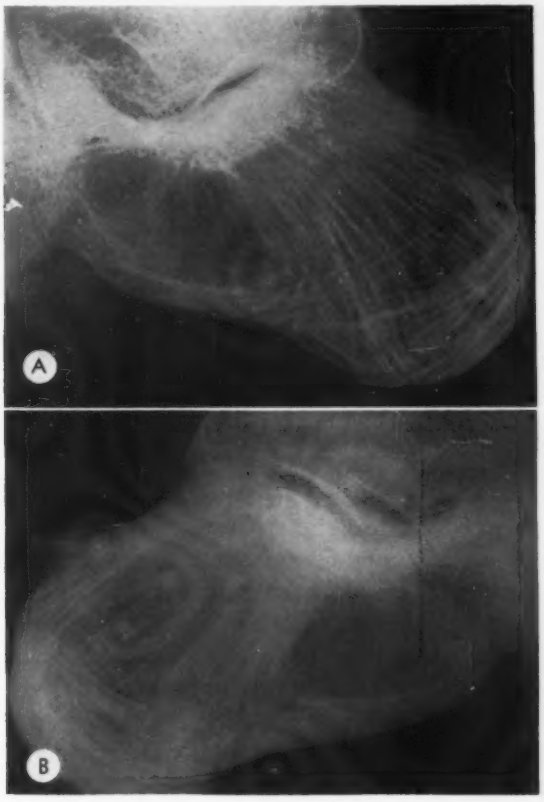

FIG. 9. Lateral roentgenograms of the right and left calcanei from the same patient. (*A*) Early active stage. Thick bundles organized along lines of stress and crossing at approximately right angles superimpose upon the generalized radiolucent bone. (*B*) The disease approaches the healed or quiescent stage.

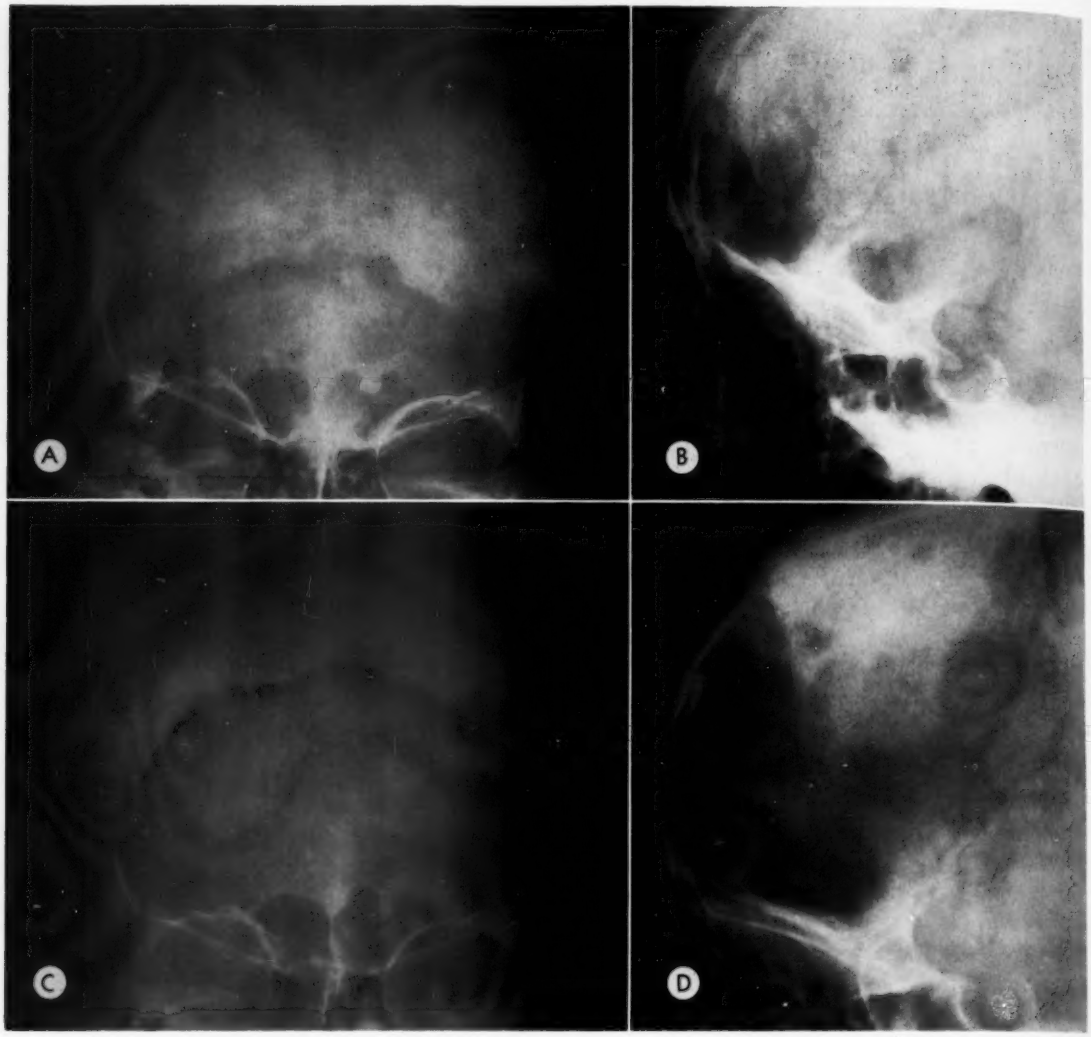

FIG. 10. (*A* and *B*) Osteoporosis circumscripta of the frontal bone. (*C* and *D*) Approximately five years later. The advancing margin is sharply demarcated from the normal bone. The meningeal vascular grooves are not obliterated as the meningeal surface of the inner table has not been destroyed.

decrease in the size of the marrow spaces; the surrounding cortex is thick and the bone is increased in size (Fig. 8). Sclerosis is seen frequently in the pelvis, vertebrae, clavicles, facial bones, ends of the long bones, base of the skull, and bones of the wrists, hands, and feet. The ribs, not commonly involved, also may be sclerotic.

In the long bones, the cortex is thickened and dense. The medullary cavity may be widened or narrowed, and in some cases almost obliterated (Fig. 1). The cancellous portions at the bone ends frequently have longitudinally thickened trabeculae. In some cases, sclerosis of the cortex is associated with the mixed type of involvement of the cancellous ends.

The sclerotic form represents the healed or end stages of the disease. The bones appear massive and strong, but because of the structurally inferior composition are more fragile than normal bones. Complete and fissure fractures occur but are less common than in the other more active forms of the disease. Of 27 fissure fractures reported by Dickson *et al.*,[7] 23 were in the combined form and only 4 in the sclerotic form of the disease.

Mixed Form. This form is the most common and may be present in any part of the skeleton. The homogeneous cortex is replaced by a cancellous structure consisting of thick, wavy bundles. The medullary cavity of long bones, although usually maintaining its width, may be widened or narrowed (Fig. 3). The trabeculae in the direction of the lines of stress are organized into thick tracts, frequently widely spaced, with intervening connective tissue. The tracts of bone are exaggerations of the stress trajectories seen in normal bones. The bones subjected to vertical stress, such as the vertebral bodies and the ends of the straight long bones, have exaggerated vertical tracts. In other bones where the distribution of forces is more complex, such as at the proximal end of the femur, the calcaneus, pelvis, and scapula, the thickened trajectories are arranged along the lines of stress counteracting the forces of tension and compression (Fig. 5; and 9, *A* and *B*). Thus, two main systems of trajectories crossing each other at right angles are arranged symmetrically around the neutral axis.

Radiolucent Form. This form is seen most frequently in the calvarium of the skull where it is known as osteoporosis circumscripta. It represents the destructive stage of the disease and, if followed for a sufficiently long period of time, it can be observed to change to the mixed and sclerotic forms. The initial lesion usually begins in the frontal or occipital areas, and, over a course of years, spreads slowly to involve the entire calvarium (Fig. 10, *A–D*). The process is usually continuous, extending almost equally to both sides, but it may involve only one side, or arise as separate foci at different locations. The diploë is diminished in density and obliterated or converted to a mesh of fine trabeculae. The outer table is thinned from within. The suture lines do not hinder progress of the disease and are eventually obliterated. The interface between diseased and normal bone is sharply demarcated with no evidence of reactive sclerosis. In time, sclerotic deposits of bone are formed at the sites which were involved earliest and may advance in the same direction and rate as the preceding destructive process. In some skulls, the normal bone, zones of radiolucency, and zones of beginning sclerosis are adjacent to each other. The sclerosis is greatest near the inner table where it may form a continuous layer adjacent to the brain. Other irregular patches appearing in the diploë are in sharp contrast to the surrounding

Fig. 11. Predominantly destructive Paget's disease with some patches of sclerosis in the frontal bone. The thickened calvarium is defined peripherally by a thin layer of periosteal bone. Basilar impression has occurred.

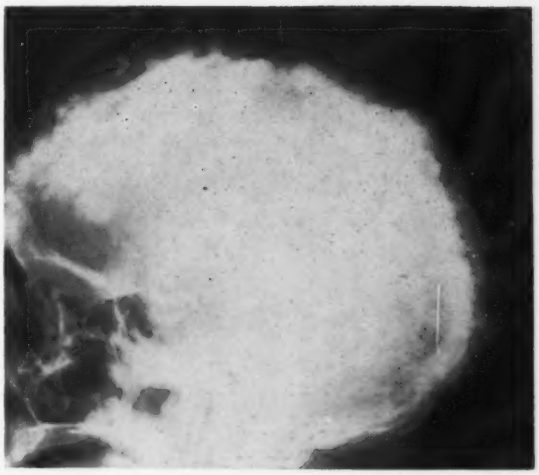

Fig. 12. Dense areas of sclerotic bone scattered throughout the calvarium are more prominent along the inner surface.

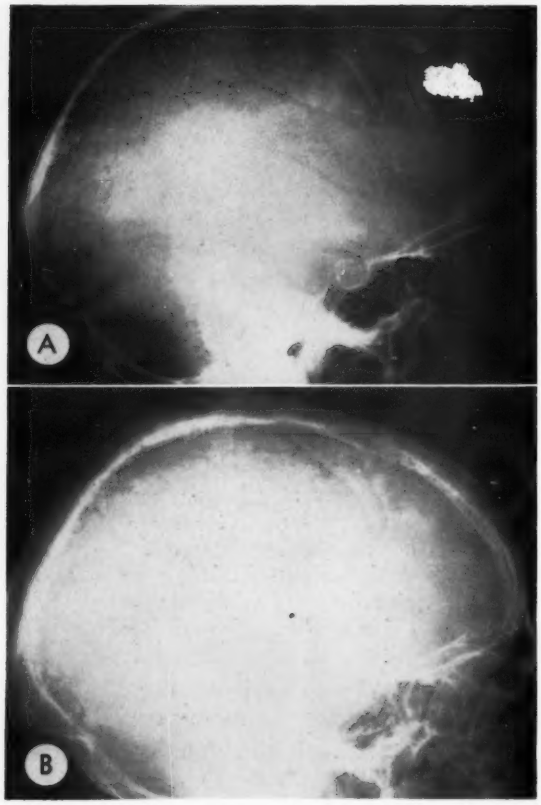

Fig. 13. (*A*) Diffuse demineralization of the calvarium associated with large meningeal grooves. (*B*) Under-exposed roentgenogram to show the smooth thin outer table and slightly thickened calvarium. The inner table is relatively dense.

radiolucent fibrous tissue. At this time the outer table migrates peripherally as a result of bone apposition on the periosteal surface and attrition on the diploic surface, resulting in a markedly thickened cranium. The thin outer subperiosteal layer is always present, although it may not be visible on the roentgenogram unless a bright light is used to view it, or unless the film has been underexposed (Fig. 11; and 13*B*).

As the destructive process subsides, new bone forms on the inner surface of the external bony plate, eventually replacing the connective tissue and producing a thick, dense outer table. The disease is then quiescent and no further alterations occur (Fig. 12).

Occasionally, the entire calvarium may be involved by the destructive process with no evidence of new bone formation visible except for a thickening of the calvarium from periosteal proliferation. The appearance may be similar to that of osteoporosis, or may differ only in the greater thickness of the skull. Some cases present a mottled radiolucency.[7] In 1 case of diffuse demineralization, the grooves for the meningeal vessels were large, possibly the result of large vessels feeding the vascular diseased bone (Fig. 13*A*).

In the long bones, extensive destructive lesions without evidence of repair is not common. Cases have been reported in which a cyst-like lesion involved the anterior superior part of the tibia[3,4,21] or other long bones. These lesions are initially spindle shaped with the cancellous bone reduced in

Fig. 14. (*A*) Normal left tibia. (*B*) Destructive lesion involving the anterior compact bone and the spongiosa of the upper third of the right tibia. The compact bone is less dense and the almost homogeneous shadow is replaced by vertical striations, presumably the result of widening of the haversian spaces. The lower border is sharply demarcated from normal compact bone in the shape of a V (arrows). The vertical trabeculae of the spongy bone are thickened.

density and the homogeneous compacta being replaced by parallel layers of spongy bone. The advancing margin is surrounded by normal bone, clearly defined in the form of a V (Fig. 14, *A* and *B*). The outer surface may be a thin layer of bone deposited by the elevated periosteum. As the lesion grows, it retains its V-shape at the periphery. This configuration differentiates it from other types of lytic lesions.

Paget's disease begins at one end or both ends of a long bone and progresses toward the other end. By studying the advancing edge of the lesion on roentgenograms, one can reconstruct the sequence of events leading from the active destructive lesions to the thick, sclerotic, healed phase. Immediately adjacent to the normal bone is an area of diminished density involving, predominantly, the center of the cortex. Longitudinal striations representing enlarged haversian canals are present (Fig. 15).

Fig. 15. Lateral roentgenogram of the femur. The destructive, sharply demarcated lesion is seen best in the posterior cortex. Vertical striations are now visible in the cortex.

Fig. 16. Active, progressing lesion of the distal tibia. The margin is sharply demarcated in V shape. The periosteum has formed a thin layer of bone, expanding the shaft. The older lesion is situated proximally. All stages in the development of Paget's disease can be seen with the sclerotic healing phase visible at the top of the roentgenogram. More proximal in the tibia, but not illustrated, the radiolucent tissue beneath the thin periosteal layer has been replaced by dense Paget's bone.

Schmorl[20] observed that this was the result of osteoclastic resorption of the haversian canal walls and their subsequent replacement by vascular connective tissue. By observing the adjacent, more extensively involved bone, it is apparent that, as the lesions approach the periosteum, a thin layer of periosteal lamellar bone is formed and expansion occurs. A bright viewing light is sometimes necessary to see this newly formed bone on the roentgenogram. The radiolucent marginal lesion and elevated periosteal new bone are indicative of still active and progressive lesions. With healing, as seen farther along the diseased

FIG. 17. This patient had bilateral symmetric fractures involving the lateral cortex of the proximal femoral shafts, seen on earlier roentgenograms. A residual scar was present in the left femur and the fracture in the right femur has some beginning endosteal and periosteal callus. Due to the fact that the bones were demineralized and had bilateral symmetric horizontal defects, and that the patient had an elevated serum alkaline phosphatase, she was treated unsuccessfully for osteomalacia. It is believed that this disease can be differentiated from osteomalacia because (1) the cortical defects involve the lateral surface of the femur, the usual site in Paget's disease but not in osteomalacia; and (2) the trabeculae along the lines of stress are thickened in the left femur.

shaft, the fibrous tissue becomes converted to bone; the periosteal bone thickens from within and ultimately is incorporated into the thickened cortex (Fig. 16).

Some cases present a generalized demineralization of a bone indistinguishable from that of osteoporosis. The trabeculae along the lines of stress thicken and form prominent trajectories which are usually more pronounced in Paget's disease (Fig. 17) than in osteoporosis, although in osteoporosis, too, the trabeculae may be thickened along the stress lines. Unless roentgenograms of other parts of the skeleton have been obtained, uncertainty about which of the two diseases is present may arise. Paget's disease, unlike osteoporosis, ordinarily involves only a part of a bone or a limited part of the skeleton (Fig. 18, *A* and *B*). Both diseases, however, occur in elderly people and are frequently associated with each other. We have had 2 patients with Paget's disease, both of whom also had thinning of the parietal bones of the skull, a condition probably related to osteoporosis.[26]

Vertebrae involved by lytic lesions may resemble hemangiomas because of the presence of thick vertical trabeculae. In these cases, similar involvement of the neural arch, spinous process, and transverse processes, or of other bones, is usually present, a condition not seen in hemangioma. The destruction of bone may be so extensive that no trabeculae will be visible within the vertebral body (Fig. 19, *A* and *B*).

The diffusely demineralized bones of the lower extremity, without thickening of the cortex, may also resemble those in osteoporosis. One patient with such diffuse demineralization of the pelvis and femora

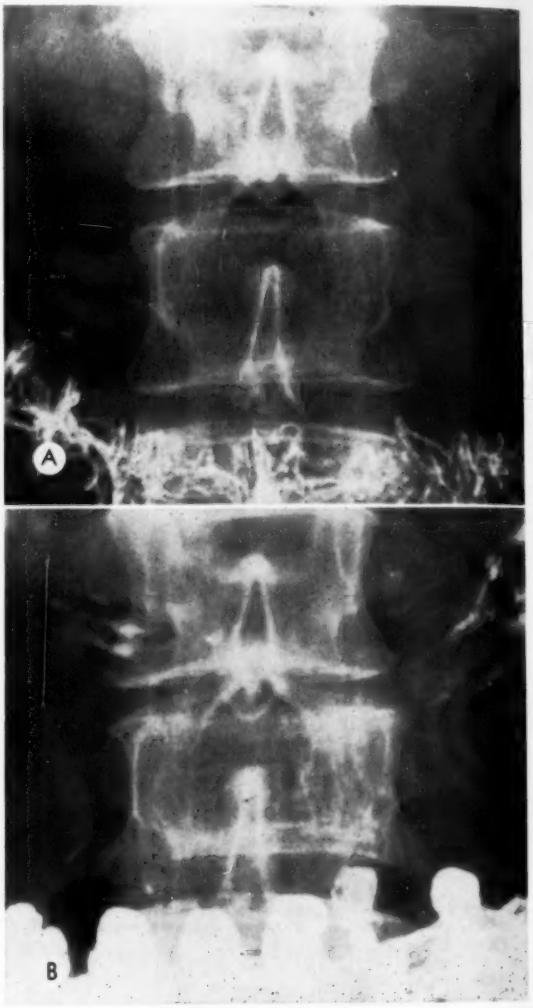

FIG. 18. (*A*) The third lumbar vertebra is less dense than the adjacent vertebrae. The pedicles are partially obscured, particularly on the left. (*B*) Three and one-half years later the typical coarse vertical striations of Paget's disease have appeared.

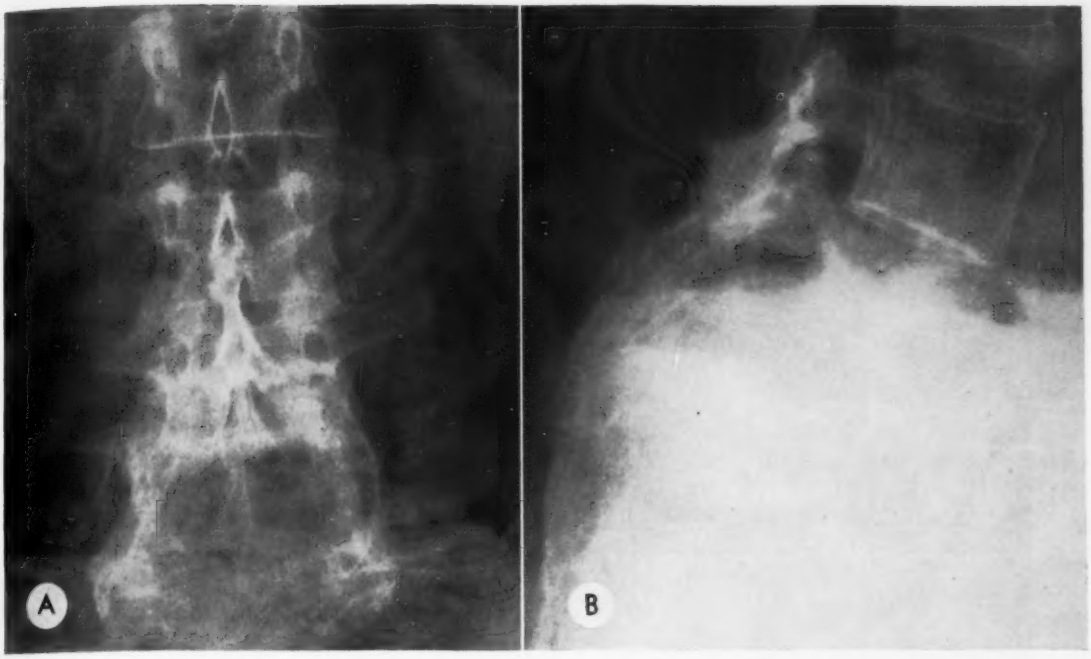

Fig. 19. (*A* and *B*) There is almost complete destruction of the fourth and fifth vertebral bodies; this had been noted for several years, diagnosed as hemangioma, and treated with roentgen irradiation. The presence of typical Paget's disease in the neural arches, attached processes, and in the sacrum, suggesting that the destruction was due to this disease, was substantiated by biopsy.

sustained a fracture of the neck of the right femur; bone biopsy revealed Paget's disease. Roentgenograms of the femur after open reduction showed the typical horizontal defects of the lateral cortical surface representing healed fissure fractures (Fig. 20, *A* and *B*). These lesions, when present, establish the diagnosis of Paget's disease.

A severe osteolytic form may occur, frequently involving only one tubular bone, in which the bone is rapidly destroyed, even the cortex disappearing in some areas. Expansion and deformity develop with striations extending in many directions (Fig. 3).

It has been stated that the articular cartilage is not involved by Paget's disease.[4,7,20] However, in cases with unilateral involvement of the pelvis or of the head of one femur, the articular cartilage adjacent to the bony abnormality frequently is narrow (Fig. 5). On the roentgenogram, the cartilage narrowing usually appears uniform throughout and is frequently not associated with hypertrophic changes. It is possible that the narrowing results from accelerated endochondral ossification with enlargement of the bone and replacement of part of the deeper layers of cartilage by Paget's bone. If this is true it could contribute to the elongation of tubular bones. Schmorl[20] observed a similar narrowing between the intervertebral disks and adjacent involved vertebrae and at the margins of the costal cartilage in Paget's disease as well as in osteomalacia and hyperparathyroidism. Others[13,24] observed the same phenomenon in acromegaly, where articular cartilage proliferation is associated with subchondral new bone formation.

Jaws and Teeth. Contrary to the opinion of some early investigators,[18,20] the disease frequently is present in the jaws.[22] Stafne[22] found it at this site in 23 of 138 cases of Paget's disease. The maxilla was involved in 20 and the mandible in 3. In 19 of the 23 cases, the skull also showed abnormality.[22]

The earliest change observed is a demineralization, which may be uneven, of the

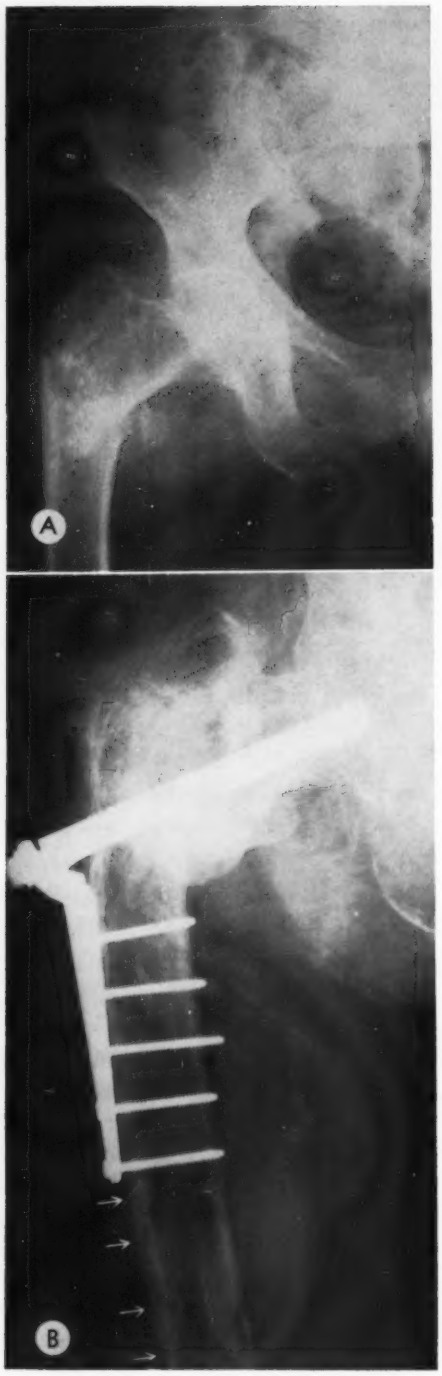

FIG. 20. (*A*) Intertrochanteric fracture of the right femur associated with diffuse demineralization of the pelvis and both femora. Slight thickening of the stress trajectories, but this is no greater than that seen in some cases of osteoporosis. A biopsy of the fracture site at the time of open reduction showed Paget's disease. (*B*) Roentgenogram obtained after surgery includes the mid-shaft of the femur, which was not seen on the preoperative roentgenograms. Typical healed Paget's clefts are visible in the lateral cortex below the metallic plate.

bone around the teeth. The trabeculae bearing the stresses of the teeth may become thickened giving a fibrillar pattern. The lamina dura disappears and the cementum and dentine about the apices of the teeth are resorbed.[6] Since the cementum is a thin shell of modified bone tissue covering the dentine of the root and neck of a tooth, it is not surprising that it also is destroyed by a disease affecting adjacent bone.[16] In some cases, the radiolucent lesions about the roots of the teeth resemble apical abscesses but the teeth are not carious and show no evidence of infection (Fig. 21). In later stages the bone becomes sclerotic, as in other parts of the skeleton, and the lamina dura does not reappear. Hypercementosis replaces the resorptive lesion, producing a bulbous enlargement of the root of the tooth which may be ankylosed to the surrounding bone.

SUMMARY

Paget's disease is primarily one of bone destruction. In the reparative phase, excess bone is formed which produces a characteristic roentgen appearance. In some cases, the long, persistent, destructive phase presents problems in roentgen diagnosis, particularly if the disease is diffuse and is present in patients who also have osteoporosis. The destructive phase of the disease can be diagnosed if: (1) the characteristic fissure fractures or their residual clefts are present in the long bones of the lower extremities; (2) the margin in compact bone can be seen to be sharply demarcated and, sometimes, associated with a thin layer of subperiosteal new bone; or (3) the trajectories formed along the lines of stress are thickened. Characteristic destructive lesions frequently are seen in the skull and about the roots of the teeth.

Bone apposition on the convex surface or

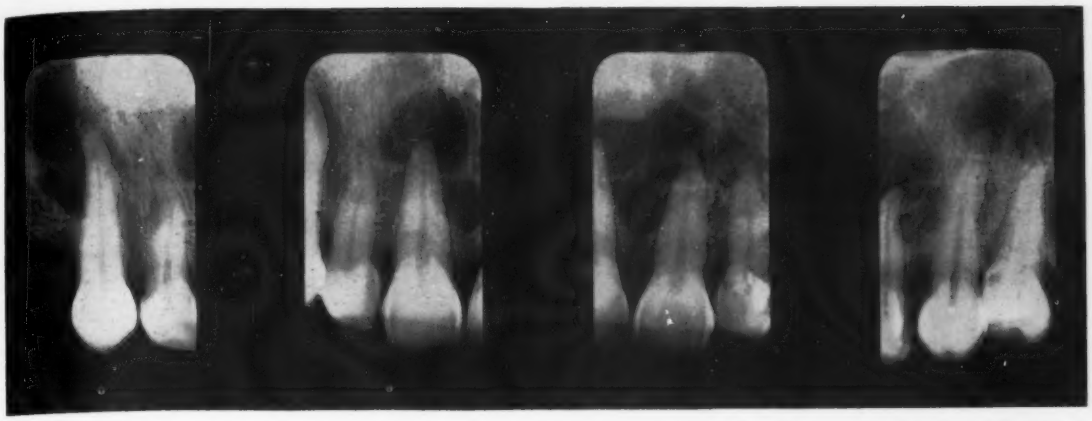

Fig. 21. Sharply demarcated destructive lesions about the roots of several upper teeth. Thick trabeculae of the surrounding bone. (Reproduced with permission of the Year Book Publishers, from "Endocrinology in Clinical Practice," 1953, p. 98.)

the surface subjected to tensile forces produces thickening of a bone. This may be followed by resorption resulting in bending of the bone without reducing its length.

The activity of the disease can be assessed by its roentgen appearance.

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EARLY CHANGES IN CHARCOT'S JOINTS*

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THE roentgen appearance of advanced Charcot's arthropathy has been well documented and presents little difficulty in diagnosis. However, the changes observed in the incipient stages of this lesion may constitute a problem in diagnosis for the radiologist and the clinician. The difficulties in the diagnosis of the evolving Charcot's joint become self-evident when it is realized that tabetic arthropathy may develop as a presenting symptom before other signs and symptoms of disease of the central nervous system are present. Twenty per cent of patients with Charcot's joints show no additional signs of cranial nerve or spinal cord disease, while serologic examinations of the blood and cerebrospinal fluid are negative in approximately 50 per cent of patients with tabes dorsalis.[15] Since treatment may be effective in arresting progress of the destructive joint lesions, recognition of the process in its early stages should contribute to more satisfactory functional salvage. Earlier recognition, as in any disease, will often spare the patient painful and usually fruitless procedures undertaken as therapeutic measures or for the purpose of establishing a diagnosis.

This paper will discuss the roentgen findings in early Charcot's arthropathy as noted in 6 cases reviewed by the authors. Although the diagnosis of neurosyphilis was established in all but 1 of these patients, the roentgen studies were misleading.

PATHOGENESIS

The actual causative factors in the development of the articular lesions are not established. Charcot believed that the etiologic mechanism was either damage to trophic centers in the spinal cord or degeneration of the peripheral nerves. Volkmann and Virchow, on the contrary, regarded trauma as the major exciting cause. Eloesser,[2] seeking to evaluate trauma as the primary force, demonstrated that typical neuropathic joint disease did not develop if only the dorsal roots in laboratory animals were severed. If, however, the anesthetized joints were subjected to direct trauma, neuropathic joint lesions were produced.

It is often stated that the Charcot joint is painless.[1] However, during the early phase of joint effusion pain is frequently present.[9] In a study designed to explain the paradox of pain in the anesthetic joint, Eloesser[2] thrust pins into experimentally produced Charcot's joints in cats. Observing that the soft tissues around the joint were extremely sensitive, while the bony surfaces and periostium were not, he concluded that soft tissue distention by the effusion was the factor which produced pain.

EARLY ROENTGEN FINDINGS

In the early stages of the disease, the roentgen findings have been reported as those of joint effusion, subluxation, various forms of degenerative bone changes and spontaneous fractures.[1,11,14] Each change may exist separately or in association with others. There is no definite chronologic order of appearance. Since severe Charcot's arthropathy may develop within several weeks, roentgen studies may not be done early enough to record incipient changes.[5] However, most cases progress slowly and in these the roentgen signs of early lesions are most apt to be observed; the roentgen interpretation must be accurate inasmuch as it may provide the only clue to the presence of the process.

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JOINT EFFUSION

Joint effusion is a common early change.[1,10,13,14] It may occur spontaneously or following a slight trauma and it varies from a mild hydroarthrosis to one of massive proportions (Fig. 1). Distention of the joint with separation of the articular surfaces is seen. The bony contours remain regular and well defined. Aspiration of the fluid is invariably followed by recurrence. A characteristic of this effusion is its chronicity and persistence.[10] The effusion is attributed to simple transudation from the vessels.[12] Trauma, enhanced by impaired sensory perception, may result in hemorrhage into the joint, adding to the volume of an effusion.

An actual increase in the mucin content of the synovial fluid may be found. This is associated with proliferation of the synovial cells and is followed by hyperplasia and thickening of the joint capsule. The capsule may become extremely thick and bulky, retaining a large amount of fluid. Capsular rupture may ensue.[6]

CASE I. A fifty-two year old Negro female was hospitalized because of pain, numbness and tingling of the lower extremities and difficulty in walking of eighteen years' duration. Approximately twenty-five years earlier she had been treated for syphilis. Neurologic examination revealed tabes dorsalis and serologic studies of the blood and spinal fluid were positive for syphilis. There was swelling and crepitation of the right knee joint.

The initial roentgenogram was interpreted as showing minimal "hypertrophic" changes and extensive joint effusion (Fig. 1*A*). Two days later there was marked increase in the effusion and enormous distention of the joint. The patella was markedly displaced by the effusion and there was slight medial subluxation of the femur on the tibia (Fig. 1, *B* and *C*). Re-examination five years later demonstrated a typical, advanced Charcot's joint (Fig. 1*D*). The patient had refused arthrodesis when first seen and had not returned for further treatment.

Comment. Although the clinical findings in this patient were readily recognized as those of tabes dorsalis, the roentgen changes were interpreted as those of hypertrophic osteoarthritis. The significance of marked effusion into the joint was not appreciated.

SUBLUXATION

Subluxation is probably related to generalized muscular hypotonia. With loss of synergistic reflexes and sensory loss, the muscles and ligaments remain the sole factors in the maintenance of joint alignment and stability. When they become overstretched, subluxation takes place. Effusion also contributes to the development of subluxation by stretching the ligaments, which become increasingly relaxed. Abnormal joint alignment takes place (Fig. 1, *B* and *C*). This in turn results in faulty weight bearing with excessive stress upon the articular surfaces and is followed by destructive changes.

CASE II. A fifty-three year old white male was treated for tabes dorsalis in 1939. Roentgenograms of the left knee revealed mild joint effusion with no abnormality of the bony architecture or joint surfaces (Fig. 2*A*). Seven months later there was a decrease in the height of the joint space at the medial aspect, sclerosis of the tibial articular surface, and increased joint effusion (Fig. 2, *B* and *C*). Fourteen months later there was further loss in stature of the joint space and increased sclerosis of the articular surfaces of the tibia and femur; amorphous, dense, calcific debris representing typical advanced disease were present in the joint space and the joint effusion had become even greater (Fig. 2*D*).

Comment. This case illustrates the more slowly developing type of Charcot's joint. Here the lesion in its early evolution cannot be distinguished roentgenologically from hypertrophic osteoarthritis.

BONE CHANGES

The osseous changes in the neuropathic joint are similar, pathologically, to hypertrophic osteoarthritis. In fact, some authors even deny the existence of a true neuropathic joint, maintaining that this is but a severe form of osteoarthritis.[14] Since Charcot's joint occurs in those age groups in which hypertrophic osteoarthritis is also widely prevalent, the differentiation of the lesions may be very difficult. The problem is

Fig. 1. Case 1. (*A*) The initial roentgenogram shows minimal "hypertrophic" changes and extensive joint effusion. (*B* and *C*) Two days after *A* there is marked increase in the effusion with enormous distention of the joint. The patella has been markedly displaced by the effusion and there is slight medial subluxation of the femur on the tibia. (*D*) Typical advanced Charcot's joint five years later.

A
B
C
D

further complicated by the fact that both diseases may be present simultaneously.

Microscopically, also, neurotrophic changes resemble those of osteoarthritis. There is degeneration and partial disappearance of the articular cartilage. The collagenous fibers are exposed and show fibrillation. Granulation tissue in the form of a pannus grows over the articular cartilage, eventually invades it, and begins to absorb it. In the neuropathic joint this pannus is more active and profuse than it is in osteoarthritis. When areas of cartilage are fully destroyed, the exposed bone begins to proliferate and produce new bone. This is a reactivation of the process of endochondral bone formation which takes place in growing bone, and it accounts for the roentgen finding of sclerosis beneath surface defects. At the periphery of the joint this active bone formation gives rise to marginal osteophytes.[5,9]

Horwitz[5] observed that bone and cartilage debris are ground into the synovial membrane early in the evolution of Charcot's joint. This feature of the microscopic pathology of the disease enabled him to establish the diagnosis in 5 patients in whom there were no definite clinical or roentgen findings of neuroarthropathy. Similar microscopic findings were present in 19 cases of advanced tabetic arthropathy, whereas in a control series of 30 joints involved by other types of arthritis there were only 2 instances in which bone and cartilage debris were found in the synovial membrane, both of the latter being far advanced osteoarthritic lesions. Horwitz[5] thus concluded that this histologic finding indicated neuroarthropathy, and was particularly useful in recognition of the early lesion and its differentiation from other types of joint disease.

Roentgenologically, diffuse sclerosis may be the earliest finding (Fig. 5*A*). The bony trabeculae are obscured and a slight increase in bone density is evident.[3,7] These changes are attributed to deposition of calcium within the interstices of the bone.[1] When the articular cartilage is destroyed, irregularities and erosions of the bony cortex are seen (Fig. 3*A* and 4*A*). Irregular, patchy areas of sclerosis may then become evident. The early osteophytic formations in Charcot's joint are said to differ from osteoarthritic spurs only by their unsharp outlines and rounded fuzzy margins, which tend to merge with the surrounding soft tissues.[6] They present a somewhat bizarre appearance and may attain enormous size and break off.

Case III. A fifty-seven old white male was hospitalized because of an incapacitating limp of the lower extremity and pain in the left hip. Five years previously he had been treated for syphilis. Serologic studies of the blood and spinal fluid were consistently negative for syphilis, although physical examination showed tabes dorsalis. Roentgen study on admission revealed moderate sclerosis of the acetabulum and an early destructive lesion of the head of the right femur (Fig. 3*A*). The left hip was already extensively destroyed, presenting typical roentgen findings of Charcot's arthropathy. Re-examination during a second admission five months later showed advanced destruction of the right femoral head characteristic of late Charcot's neuroarthropathy (Fig. 3*B*).

Comment. The lesion in the right hip, representing incipient Charcot's disease, was indistinguishable in its roentgen appearance from hypertrophic osteoarthritis. Despite characteristic findings of severe Charcot's arthropathy in the left hip, the initial changes of a similar lesion in the right hip were regarded as "degenerative." Attention was focused on the severely damaged left hip to the neglect of the right

Fig. 2. Case II. (*A*) Roentgenogram of the left knee reveals slight joint effusion. The bony architecture and joint surfaces are normal. (*B* and *C*) Seven months later there is a decrease in the height of the medial aspect of the joint space and slight sclerosis of the tibial articular surface. The joint effusion has increased. These changes are indistinguishable from those of hypertrophic osteoarthritis. (*D*) Fourteen months later there is further reduction in the joint space and increased sclerosis of the articular surfaces of the tibia and femur. Dense amorphous calcific debris is present in the joint space indicating advanced disease. The joint effusion has also increased considerably.

FIG. 3. Case III. (*A*) Roentgen study of the right hip reveals moderate sclerosis of the acetabulum and an early sclerotic lesion in the head of the right femur. (*B*) Re-examination five months later reveals severe destruction of the femoral head. The amorphous calcification within the joint space and the dense bone sclerosis of the involved femoral head are characteristic of the late changes in Charcot's joint.

which, thereafter, became almost totally destroyed within a five month period. The patient offered no complaints regarding his right hip. These changes illustrate the early roentgen findings in the evolving Charcot's joint.

CASE IV. A fifty-six year old white male was hospitalized for treatment of hypertensive heart disease and congestive heart failure. Diagnostic studies included a roentgenogram of the abdomen which revealed an abnormality of the right hip. Further roentgen study disclosed narrowing of the joint space, sclerosis of the acetabulum and cyst-like areas of radiolucency and mottled areas of sclerosis producing deformity of the femoral head and neck (Fig. 4*A*). The findings were interpreted as those of osteoarthritis. The patient had no complaints referable to the right hip and he was discharged after recovery from his congestive heart failure.

Re-examination four months later, when the patient was readmitted primarily for a complaint of pain in the right hip area and weakness of the right lower extremity, revealed advanced destruction of the femoral head and considerable calcific debris in the distended joint space (Fig. 4*B*). On the basis of these roentgen findings the diagnosis of Charcot's joint was suggested, although there was no supporting evidence in the history, physical findings or laboratory studies. Numerous examinations of the blood and spinal fluid and repeated neurologic examinations failed to provide evidence of syphilis. In an effort to establish a diagnosis and rule out neoplasm, two surgical biopsies were performed but neither revealed the true nature of the lesion.

Two years later the patient had a spontaneous fracture at the proximal intercondylar portion of the right tibia. This was entirely unrelated to trauma or sprain, was painless and was manifested by severe ecchymosis of the leg and knee. Until roentgenograms disclosed the fracture, the lesion was diagnosed as a "local subcutaneous hemorrhage probably due to blood dyscrasia." Finally, after a period of more than two years from onset of the articular lesions, the diagnosis of syphilis in this patient was established by tests for *Treponema pallidum*.

Comment. This case illustrates the elusive nature of the diagnosis of Charcot's joint in its incipient phase. Early, the lesion simulated the commonly observed roentgen findings of hypertrophic osteoarthritis. Later, even characteristic roentgen findings of neuropathic joint disease were not recognized because of lack of historical, clinical or laboratory evidence of syphilis. Finally, a spontaneous fracture was interpreted clinically as a local hemorrhagic

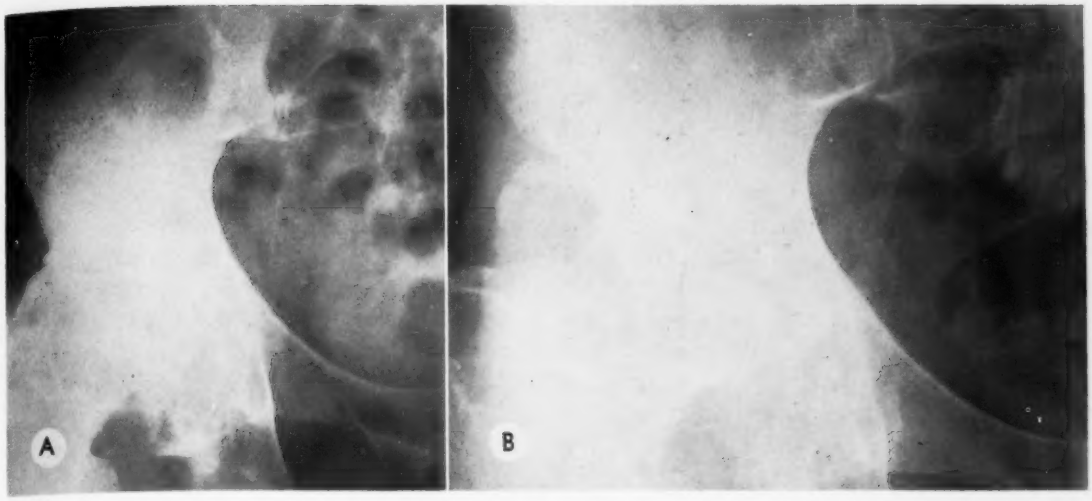

Fig. 4. Case iv. (*A*) Roentgenogram of the right hip shows narrowing of the joint space, sclerosis of the acetabulum and cyst-like areas of radiolucency with mottled areas of sclerosis producing deformity of the femoral head and neck. These changes were interpreted as due to osteoarthritis. The patient had no complaints referable to the right hip area. (*B*) Roentgenogram of the same hip four months later, when patient was readmitted primarily for a complaint of pain in the right hip and weakness of the right lower extremity, reveals advanced destruction of the femoral head and considerable calcific debris in the distended joint space. On the basis of these findings the roentgen diagnosis of Charcot's joint was suggested although there was no supporting evidence in the history, physical findings or laboratory studies.

manifestation of a blood dyscrasia and, even when the fracture was demonstrated, it was not immediately associated with the coexisting Charcot's joint.

Case v. A fifty-six year old white male was known to have acquired syphilis in 1917 but he received no treatment until 1932, when he was given malarial therapy. In 1956 he was hospitalized because of weakness of both legs and difficulty in walking of three months' duration. Neurologic examination revealed tabes dorsalis. Laboratory studies of the blood and spinal fluid were positive for syphilis. Roentgenographic study revealed marked narrowing of the disk space between L_2 and L_3 with sclerosis of the proximal portion of the body of L_3 and the adjacent portion of L_2 (Fig. 5*A*). Two and one-half years later there was marked destruction of the apposing surfaces of the vertebral bodies with beginning development of large osteophytes (Fig. 5, *B* and *C*).

Comment. Despite recognition of the classic clinical signs and symptoms of tabes dorsalis, the early changes in the lumbar spine were regarded as "disk injury or degeneration with secondary arthritic changes." The later changes were attributed to osteomyelitis of the spine with spinal cord involvement. As visualized in the initial study, the changes were entirely consistent with degenerative disease of the intervertebral disk. However, the roentgen appearance of the lesion two and one-half years later, while by itself suggestive of infectious process, could hardly have been interpreted as such in view of the earlier studies which in no manner suggested incipient spinal osteomyelitis.

Spontaneous Fractures

Charcot himself first noted the frequency of spontaneous fractures in tabes dorsalis. Sudden fracture is often the first sign of the disease.[13] A fracture may occur without preceding articular changes and prior to the onset of neurologic manifestations.[1,11,14] Typically, these fractures occur in or near a joint with predilection for the femur; the tibia, fibula, radius and ulna follow in order of decreasing frequency. Characteristically, these fractures are transverse, in contrast to the oblique and spiral fractures which follow ordinary trauma (Fig. 6*A*). With good immobilization the fractures may heal well. However, adequate immobilization is usually difficult to achieve because of the neurologic lesion.[8]

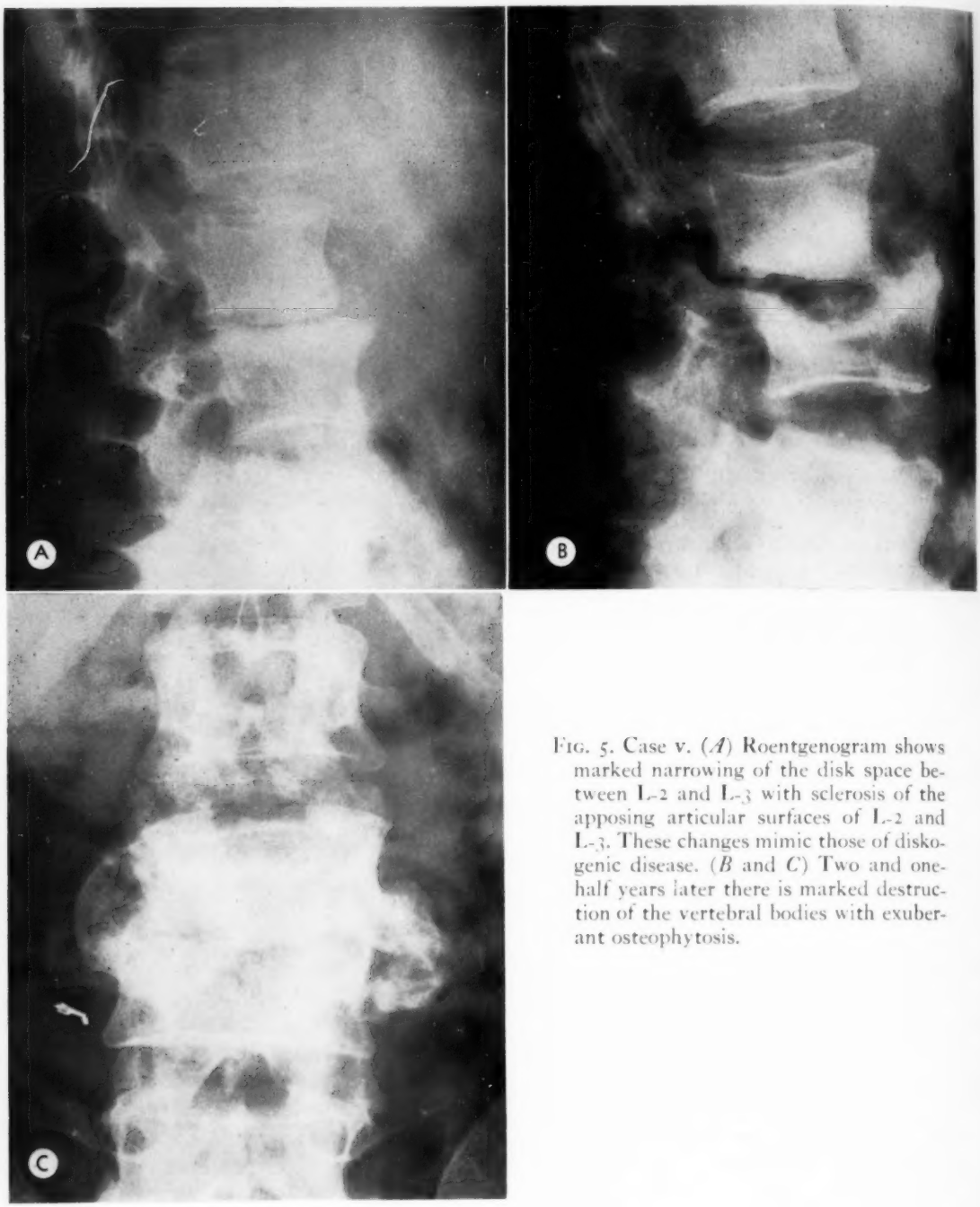

FIG. 5. Case V. (*A*) Roentgenogram shows marked narrowing of the disk space between L-2 and L-3 with sclerosis of the apposing articular surfaces of L-2 and L-3. These changes mimic those of diskogenic disease. (*B* and *C*) Two and one-half years later there is marked destruction of the vertebral bodies with exuberant osteophytosis.

Chemical analysis of the osseous material in the fracture site has demonstrated an actual increase in the inorganic salts. On the other hand, a deficiency of the organic substance has also been observed. Thus, excessive deposition of calcium salts within a poorly developed matrix results in increased brittleness of the bone.[14]

CASE VI. A fifty-six year old white male was treated for syphilis in 1952. Two years later he developed severe "shooting" pains in the lower extremities and difficulty in walking. Two

Fig. 6. Case VI. Roentgenogram made eight days following hospital admission reveals a comminuted, transverse fracture of the proximal end of the tibia with depression of the medial plateau. This type of transverse fracture, occurring spontaneously or with relatively minor trauma, is characteristic of Charcot's joint.

years after the onset of these symptoms, his right leg gave way suddenly and spontaneously and he was admitted to the hospital ten days later with a swollen and painful right knee.

Roentgen study performed after the patient had been in the hospital for eight days (eighteen days after the initial episode) disclosed a comminuted, transverse fracture of the proximal end of the tibia with depression of the medial plateau (Fig. 6). Surgical fusion was performed. Blood and spinal fluid remained positive for syphilis.

Comment. Although this patient presented with a history, physical signs and symptoms of tabes dorsalis, the swelling of the knee joint following a simple twisting injury was diagnosed as thrombophlebitis and treated with hot, moist applications and antibiotics. It was not until more than a week after admission that roentgen examination disclosed a fracture which had not been suspected clinically. Even then the fracture was not recognized as a manifestation of tabetic neuroarthropathy. During his hospital stay the lancinating pains in the right lower extremity were attributed to the fracture, although they were recognized later as a symptom of the neurologic lesion. Although this patient had had severe pains in *both* lower extremities, nevertheless there was continued reluctance to classify the fracture of the right tibia as a Charcot's joint. Even the operation for joint fusion was done primarily to control pain, an end which could not be achieved by surgical arthrodesis since the pains were of central nervous system origin.

SUMMARY

In 6 patients roentgen findings representing early changes of Charcot's neuroarthropathy were observed to be: (1) persistent joint effusion; (2) minimal subluxation; (3) degenerative changes of bone manifested by bone sclerosis, erosion and destruction which were difficult to differentiate clinically and roentgenologically from osteoarthritis; and (4) spontaneous fracture unrecognized clinically.

These changes may occur prior to any other clinical or laboratory manifestations of the disease and may masquerade as other lesions of the joints. They are particularly likely to remain unrecognized as manifestations of Charcot's neuroarthropathy.

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CERVICAL DISCOGRAPHY*

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DISCOGRAPHY is a method of introducing radiopaque material directly into a disc preceding the roentgen study. It is relatively new, the first experience being recorded in 1932 by Schmorl and Junghanns.[3] Their report was concerned with lumbar disc injections, which today are routine procedures in many parts of the country. Cervical discography, however, was not attempted until Smith and Nichols[4,5] and Cloward[1] performed it many years later.

ROENTGEN STUDY OF THE CERVICAL SPINE

Previously, roentgen study of the cervical spine was limited to routine roentgenography and visualization of the dural sac by myelography. Cervical myelography, first carried out with lipiodol and later with pantopaque, has both advantages and hazards. The advantages are these: it can be accomplished quickly by fluoroscopy and roentgenography; most of the opaque medium can be removed following the examination; it can be repeated many times; spinal cord tumors or other obstructing lesions of the spinal pathway are easily identified, and medial or lateral disc protrusions can frequently be demonstrated. The chief hazard is that the pantopaque occasionally escapes into the cranial cavity and, once it reaches the cranial subarachnoid space, it is rarely eliminated. The possibility of adverse effect on the central nervous system then may remain indefinitely. Too, this method frequently leads to both false positive and false negative delineations of extradural lesions. The neurosurgeon may consequently perform an unwarranted operation or fail to operate when it is necessary.

CLINICAL FEATURES

When one realizes the number of neurologic syndromes—from amyotrophic lateral sclerosis to hysteria—that the neck and cervical spine can mimic, the necessity of our search for another technique—neuro logic, roentgenographic, or otherwise—to assist in clarifying the diagnosis becomes apparent.

When the central nervous system is involved by neck injury or disease in such a way that the examination clearly identifies the location, extent and character of the lesion, the problem is simple. But the neurologic examination, valuable as it is, is usually inadequate by itself to establish a diagnosis that warrants surgical exploration of the neck. False negative or false positive roentgen evidence associated with indefinite neurologic findings further complicates the already difficult diagnosis.

A busy neurologist or neurosurgeon who does not thoroughly pursue the complaints of a partially or totally disabled patient who has few objective symptoms may conclude, despite the many complaints, "I can find no neurologic evidence of cervical spine or neck injury or disease." Had he pursued the patient's complaints more diligently, he might have discovered the nature of the injury or disease. This patient's pain, although it cannot be seen, is real and the muscle spasm and muscle tenderness usually accompanying it certainly indicate a disorder involving the nervous system and may indicate a neck injury or a disease and its extent.

All the structures about a cervical disc may be affected by injury or disease. Each type of injury or disease has a separate and distinct pattern of symptoms. Not only

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may the spinal cord and the peripheral nerves in the neck be affected, but the intrinsic nerve supply to the bones, ligaments, muscles and blood vessels may also be involved. When the latter occurs, these structures become painful irrespective of injury to the nearby spinal cord and peripheral nerves.

The muscle tenderness, pain and spasm frequently present with any joint injury may extend for a distance both proximal and distal to the painful joint. Since the cervical intervertebral space is a joint, being composed of an annular ligament, central nuclear structures, and cartilaginous plates upon the vertebral bodies, it too may be the source of muscle tenderness, pain and spasm.

In spite of a consistent pattern of symptoms identifying the pathologic lesion in the cervical spine, physical findings and those of routine roentgenography and even myelography may be so meager that they cast doubt as to its presence. An additional method of survey which will pinpoint the pathologic process is often necessary, and we have found it in discography.

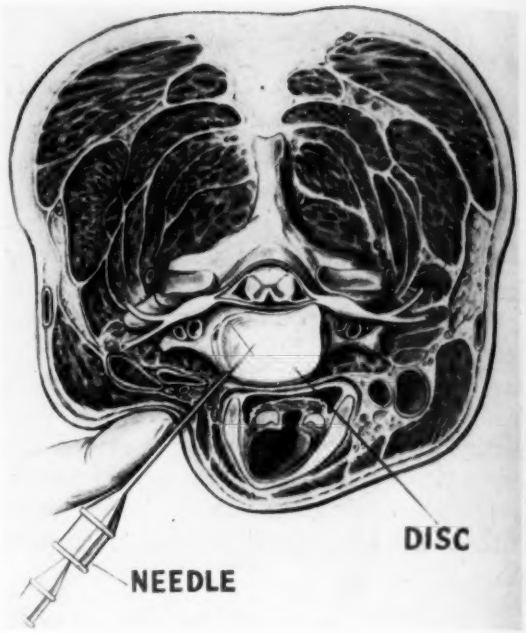

Fig. 1. Drawing showing the needle in place for discography.

Technique of Cervical Discography

Discography (Fig. 1), in contrast to myelography, is an accurate method of evaluating disc derangement. It not only reveals the disc pathology and its extent in detail but it reproduces the symptoms of the patient so that the operator may compare them with the original complaints. In contrast to any other method available at this time, it has demonstrated the highest degree of accuracy in identifying the symptomatic pathologic disc or discs.

To inject a cervical disc, a kit of equipment containing these items is used: three 20 gauge 3 inch needles for making the neck puncture, three $3\frac{1}{2}$ inch 26 gauge spinal needles to inject the disc, two 1 cc. Luer-Lok tuberculin syringes to inject the radiopaque substance into the disc, and one 2 cc. syringe with a hypodermic needle to inject the local anesthetic.

After the skin of the neck is cleansed and antiseptic has been applied, local anesthetic is injected into the skin on the side of the neck. The operator palpates the medial border of the sternomastoid muscle and the carotid artery with his left index finger; the tip of this finger is then passed beneath the trachea and esophagus until the anterior surface of the vertebral column is felt. At this point, if osteophytes are present on the anterior and anterolateral borders of the vertebrae, they can be palpated, and their presence assists in identification of the disc or discs to be injected with the opaque medium.

A needle is inserted into each disc under suspicion until the condition of that disc is ascertained; this is done by passing the 3 inch 20 gauge needle along the left index finger and through the skin and deep structures until it touches a vertebral body or the annulus of a disc. If the operator feels the point of the needle touch the vertebra, he confirms this finding by gently tapping the needle with his finger. The soft tissues are then released and are allowed to "climb back" on the needle. This encounter of the vertebral body with the needle causes the

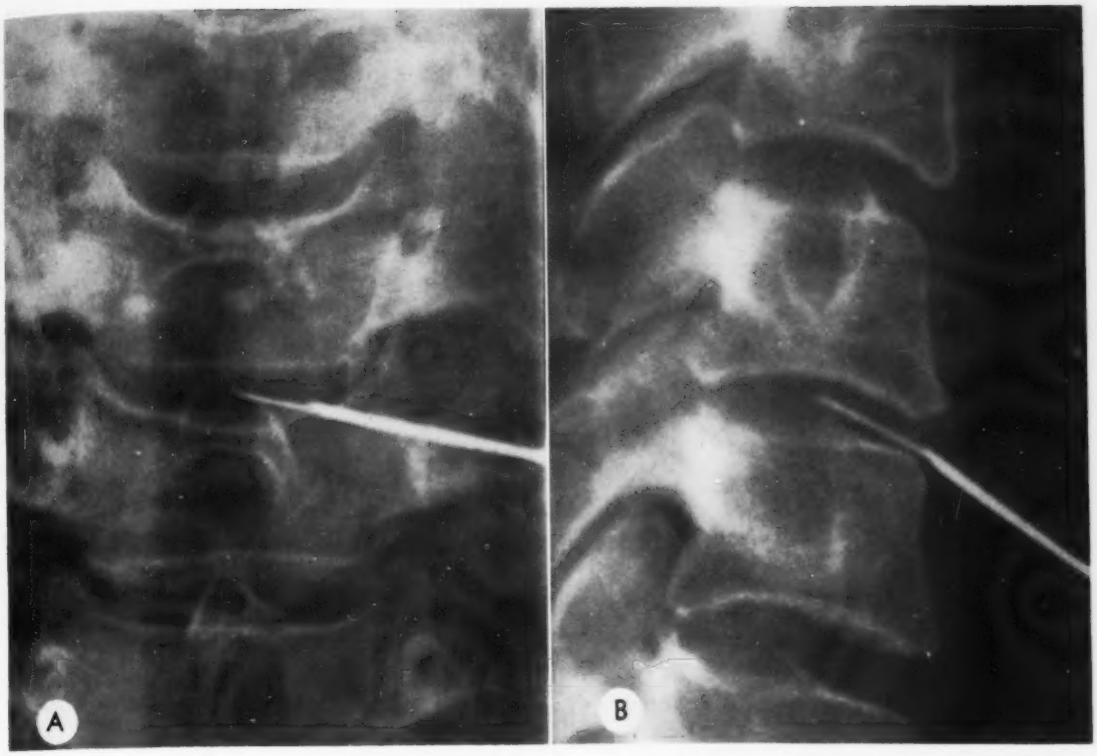

Fig. 2. (*A*) Anteroposterior and (*B*) lateral roentgenograms taken to establish the identity of the disc and the position of the needle.

patient to experience deep pain in his neck at the site of the needle. As the needle is "walked" along the vertebrae, each step causes pain locally in the neck until the annulus of a disc is encountered. Whether the annulus of the disc is encountered before or after the vertebral body, the patient experiences pain in the shoulder blade on the same side.* The operator is assured that the needle is in the annulus of the disc by the evidence of the spread of pain to the scapular area.

The 26 gauge spinal needle is then passed through the 20 gauge needle to the center of the disc. As soon as the 26 gauge needle is inserted, an anteroposterior and a lateral roentgenogram of the cervical spine are taken to establish the identity of the disc injected and the position of the point of the needle (Fig. 2, *A* and *B*). If the needle is not in the proper location, it must be shifted and another pair of roentgenograms taken. At times it may be necessary to do this repeatedly before the position is perfected.

A 1 cc. Luer-Lok tuberculin syringe is filled with 50 per cent hypaque, the stylet is removed from the 26 gauge spinal needle and the syringe is connected to it. Slowly the hypaque is injected. In a normal disc, resistance is felt after the injection of 0.2 to 0.3 cc. of hypaque and this resistance stops the injection, the patient experiencing only a moderate amount of neck and shoulder pain. But pathologically deranged discs will accept from 0.5 to 1.5 cc. of hypaque, and the patient will usually experience immediate extreme pain, numbness and weakness in the head, neck, shoulders, arms and hands.

Because this opaque medium is diffused and absorbed rapidly, the roentgenograms must be taken quickly. The regular roent-

* Cloward[2] has recently reported that the sensory nerve supply to the annulus is by way of the sinuvertebral nerves. These nerves innervate the annulus of the discs in a segmental and unilateral manner. The area of skin reference of the annulus is along the vertebral border of the scapulae.

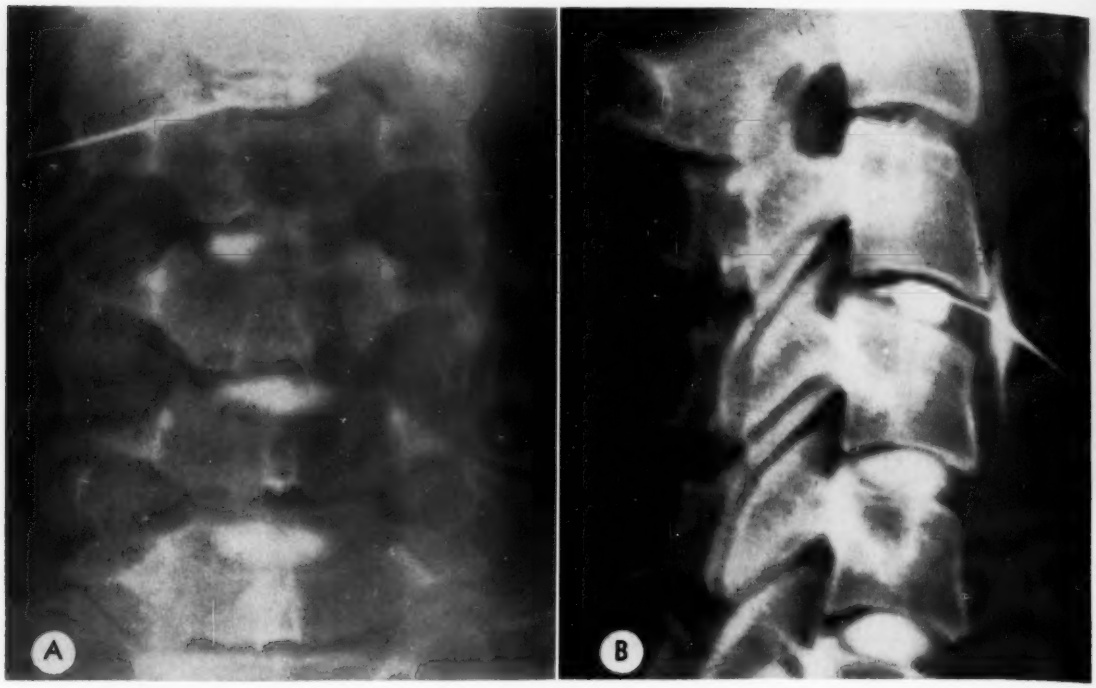

FIG. 3. (*A* and *B*) The disc may be normal with opaque medium concentrated centrally in a globule.

FIG. 4. The disc may be deranged only in an anterior portion.

genographic table with the Bucky diaphragm attached has proved satisfactory. In this case, the lateral roentgenogram is usually taken with a grid; the head unit is ideal for this work because it can be rotated about the neck.

The greatest amount of information is obtained by taking anteroposterior, lateral and oblique roentgenograms of the cervical spine. Additional disc injections should be carried out separately in order to prevent the pathologic findings on the roentgenograms from overlapping.

RESULTS

The purpose of discography is to visualize the derangement of a disc by a roentgen study of the route of the opaque medium through the disc, with particular reference to its point of concentration, the place of its extrusion through the torn annulus and its further spread into the surrounding soft tissues. When the patient experiences a recrudescence of pain, numbness and weakness during the injection, we have identified the symptomatic deranged

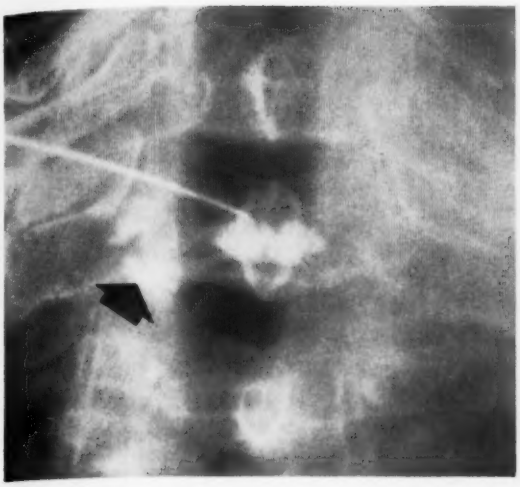

Fig. 5. The derangement may be unilateral.

disc. Damage to the disc can then be estimated and surgical therapy planned.

The discographic survey may reveal *any one* of these conditions: (1) the disc may be normal with the contrast material concentrated centrally in a globule (Fig. 3, *A* and *B*); (2) the disc may be deranged only in an anterior portion (Fig. 4); (3) the derangement may be unilateral (Fig. 5); (4) the entire disc may be severely deranged (Fig. 6); (5) the annulus of the disc may be torn loose from its attachment to the vertebral body (Fig. 7); (6) the annulus may be torn posteriorly or laterally and allow the opaque medium to spill into the adjacent soft tissues (Fig. 8); (7) disc protrusion may be present in the midline anterior

Fig. 6. The entire disc may be severely deranged.

Fig. 7. The annulus of the disc may be torn loose from its attachment to the vertebral body.

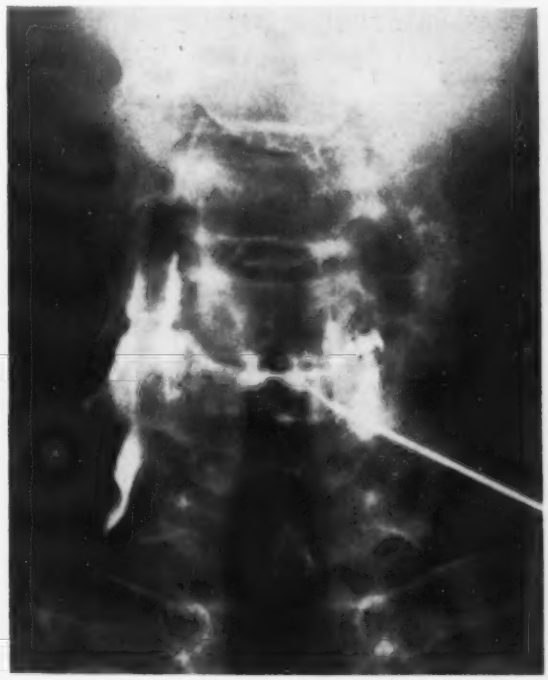

Fig. 8. The annulus may be torn posteriorly or laterally and allow the opaque medium to spill into the adjacent soft tissues.

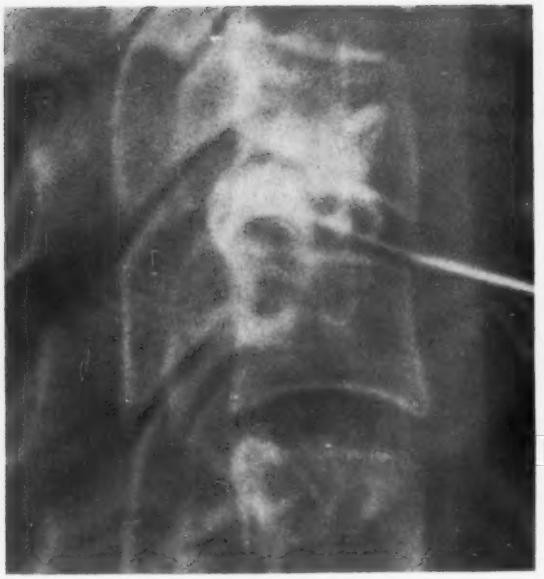

FIG. 9. Disc protrusion may be present in the midline anterior to the spinal cord.

FIG. 11. The anterior longitudinal ligament.

to the spinal cord (Fig. 9); or (8) in the oblique views disc protrusion may be seen in the nerve foramen (Fig. 10, *A* and *B*).

In addition, these anatomic structures may be identified: (1) the anterior longitudinal ligament (Fig. 11); (2) the posterior longitudinal ligament (Fig. 12); (3) the joints of Luschka (Fig. 13); (4) the vertebral cartilage plate (Fig. 14); (5) the dura and epidural space (Fig. 15); (6) the perineural space and nerve root (Fig. 16); and (7) the vertebral artery (Fig. 17, *A* and *B*).

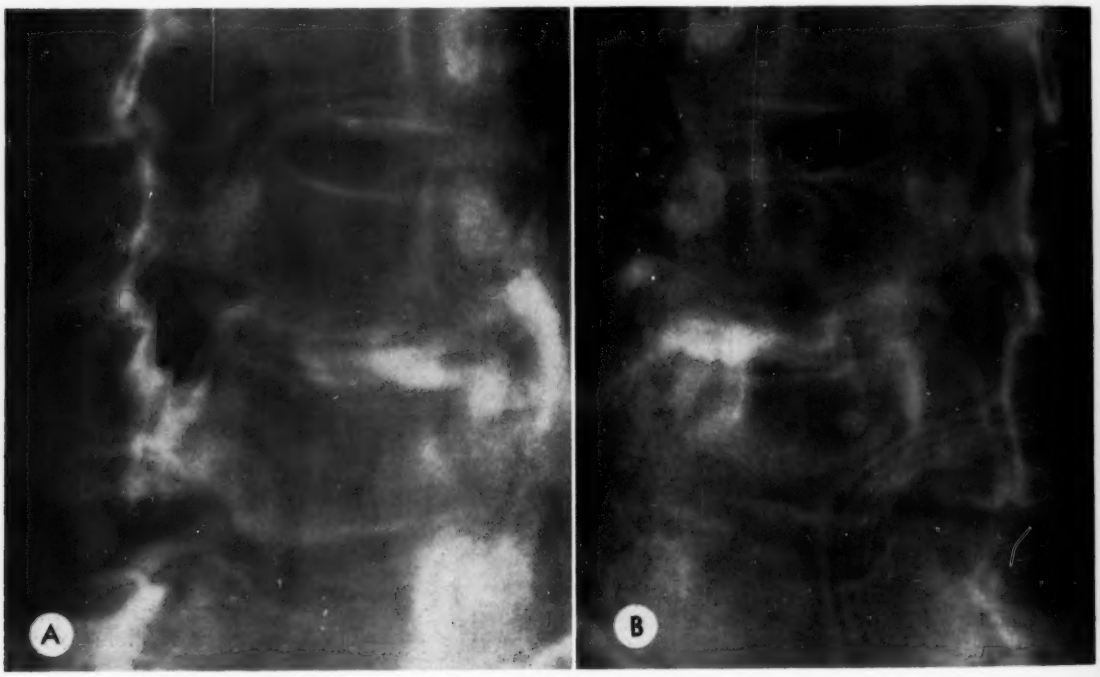

FIG. 10. (*A* and *B*) In the oblique views, disc protrusion may be seen in the nerve foramen.

Fig. 12. The posterior longitudinal ligament.

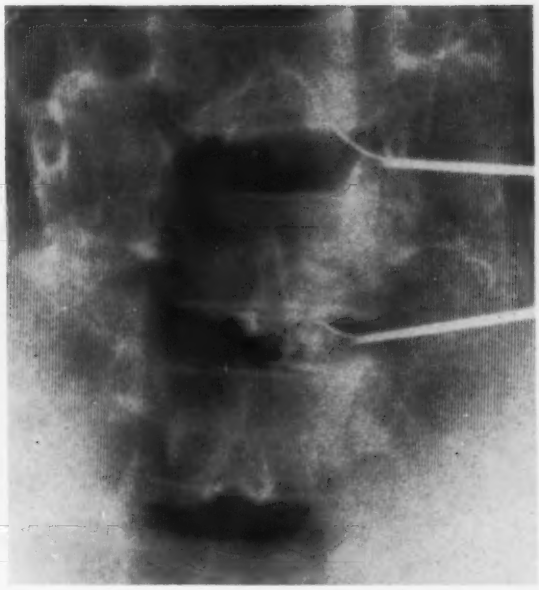

Fig. 14. The vertebral cartilage plate.

SUMMARY AND CONCLUSIONS

Discography, by demonstrating roentgenographically the internal structures of a disc, identifies both the normal disc and the deranged disc.

Cervical discography is accomplished more easily than cervical myelography and with greater safety. It delineates the normal and the deranged disc so accurately that the type and extent of disc derangement can be seen in detail and the need for disc excision can be readily established.

Fig. 13. The joints of Luschka.

Fig. 15. The dura and epidural space.

FIG. 16. The perineural space and nerve roots.

Injection of the disc irritates the nerve supply to the disc, the adjacent nerve roots and, occasionally, the nearby spinal cord and causes the patient's symptoms to be reproduced. This evidence assists the surgeon in identifying the pathologic disc needing treatment.

Cervical discography has thus proved to be a major adjunct to the routine roentgenographic study of the cervical spine.

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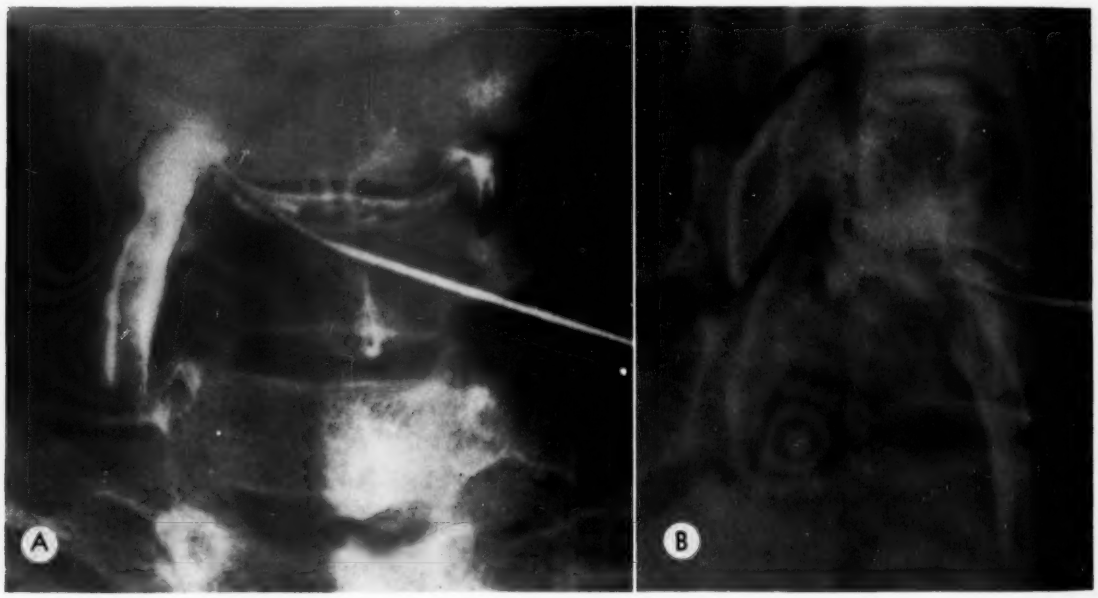

FIG. 17. (*A* and *B*) The vertebral artery.

ISOINTENSITY PATTERNS OF SCATTERED RADIATION AROUND A MIRROR OPTIC PHOTOFLUOROGRAPHIC UNIT*

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WITH the development in the last few years of the much faster mirror optic cameras for photofluorographic procedures, exposure dosages to patients have been reduced so that they more closely approach the amounts obtained from conventional chest roentgenography. These newer cameras also play an important role in the protection of technical personnel. An investigation of the scattered radiation around a modern photofluorographic unit containing a mirror optic system has been carried out; it shows a considerable reduction compared with previously published results when lens type cameras were employed.[1,2]

EQUIPMENT AND PROCEDURE

Radiation measurements were made around a 70 mm. straight hood (type X-70S) Fairchild-Odelca camera. The working aperture is f.0.8. The camera and roentgen-ray tube are supported as one single unit so that vertical movement of one results in the same movement of the other. The collimation of the primary beam was aligned by placing roentgen-ray films at the camera surface 40 inches from the target. Exposures made in this fashion permitted correction of an original malalignment and ensured that the primary beam was confined to the fluoroscopic screen surface of the camera and hood.

The position of the chest survey unit in the relatively small lead lined examination room is demonstrated diagrammatically in Figure 1. The equipment is controlled from behind a protective barrier located in one corner of the room. The tube is operated at 90 kv. peak and 150 ma. with the exposures phototimed. The roentgen-ray beam is filtered with 2.5 mm. of aluminum. The film used is Kodak Photoflure, PFG 469, green sensitive.

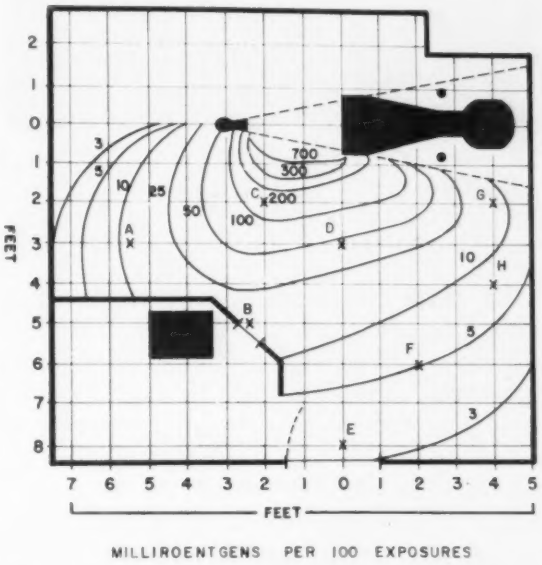

FIG. 1. Isointensity curves, indicating the distribution of scattered radiation around a mirror optic photofluorographic installation.

Measurements were made with Victoreen Model 362 pocket ionization chambers and with a .025 r Victoreen chamber. The pocket chambers were selected for nonleakage and a linear response of from 0 to 200 mr; they were found to be accurate within 2 per cent of full scale when tested with a standard radium source.

Figure 2 illustrates the arrangement of a number of chambers. The small chambers were spaced at 6 inch intervals on a portable stand. This stand was readily moved to various positions throughout the room. It allowed a large number of measurements to be made simultaneously. The active vol-

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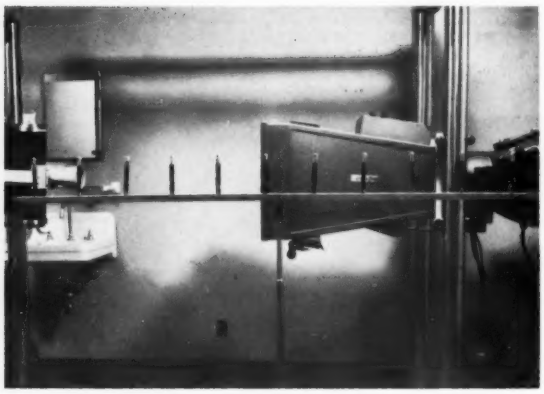

FIG. 2. An adjustable stand holds pocket ionization chambers near a 70 mm. straight hood photofluorographic unit.

umes of the chambers were adjusted to the same plane as that of the horizontal beam during each exposure. Measurements of scattered radiation were obtained both from patients and from a tissue equivalent masonite phantom. The phantom gave an exposure equivalent to that of a patient with a chest measurement of 22 cm. at the nipple line. Most of the measurements carried out for the isointensity patterns were obtained using the phantom. From measured points of equal intensity, isodose curves were constructed and are shown superimposed on the room layout (Fig. 1). The curves are expressed in milliroentgens per 100 exposures.

DISCUSSION

Morgan and Lewis[1] working with a 35 mm. camera and Van Allen[2] employing 70 mm. cameras, all of the lens type, have published isodose curves for such installations. Our isointensity patterns show a reduction in scatter of major degree. Although the phantoms that these investigators used were in each instance equivalent to a patient 2 cm. larger in chest diameter than ours, this is not considered to be a significant factor. Undoubtedly, the faster present day films do play a part in the observed differences, but the major change is related to the difference in camera speeds.

The experiments also differ in the sizes of the rooms used. Our data were accumulated in a room one-seventh the size of the rooms of the other investigators. Therefore, our measurements include backscatter from the walls to a much greater degree.

If one selects random points at equal distances from the equipment, the reduced scatter using the mirror optic camera is impressive. Table 1 compares the scatter at such points of measurement for the two types of cameras.

TABLE I

SCATTERED RADIATION USING DIFFERENT PHOTOFLUOROGRAPHIC CAMERA SYSTEMS
(in mr/100 exposures)

Points of Measurement	Lens Optics		Mirror Optics
	Morgan and Lewis[1]	Van Allen[2]	Present Authors
A	70	250	14
B	40	110	14
C	320	350	150
D	80	200	40
E	10	30	3
F	12	30	5
G	10	200	12
H	12	40	7

At only one of these points does the radiation in the mirror optics data exceed that of the lens systems. Figure 1 indicates that this point is close to a wall. It is probable that backscatter accounts for the high value.

Values for areas outside of the room and in the control section were much less than 1 mr per 100 exposures. The walls of the room and of the protective barriers around the control contain 1/16 inch of lead.

SUMMARY

Measurements made around a modern chest photofluorographic roentgen-ray installation utilizing a mirror optic system indicate less scattered radiation compared with that from lens type photofluorographic cameras.

A diagram shows a typical installation of a mirror optic type camera with its field patterns of scattered radiation.

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EXPERIMENTAL OPACIFICATION OF ORGANS AND STRUCTURES TO ROENTGEN RAYS*

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IN 1937 the authors described a method for the permanent roentgen visualization of the colon based on the infiltration of its walls with particulate opaque material.[4] This method proved useful for physiologic studies of the colon in animals.

In recent years, devices capable of subjecting animals to high acceleration or deceleration have been developed in connection with space programs, and some of the men in charge of the medical aspects of these programs have been interested in the displacement of various organs and structures during such stresses. These displacements are usually studied by strapping the animals to high velocity sleds and taking instantaneous roentgenograms at the moment of maximum deceleration. Some of these studies were not satisfactory because of insufficient roentgenographic contrast, and the authors were requested to supply a number of dogs in which organs had been made opaque to roentgen rays. Therefore, the purpose of the following experiment is somewhat unique, but the methods employed are susceptible of ordinary physiologic applications and are reported for this reason.

MATERIAL AND METHOD

Thirty-one dogs were prepared. The organs opacified were the liver, spleen, colon, stomach, duodenum, short loops of small bowel, urinary bladder, kidneys, diaphragms, and the pericardium.

Visualization of the liver and spleen was achieved by the intravenous administration of thorotrast. All other procedures required opening the abdominal or thoracic cavity under general anesthesia. After isolation of the organ or structure to be visualized, a suspension of a particulated sterile opaque material was introduced just under the outer surface by using a No. 26 needle to make multiple small injections about 1 cm. apart. Following this, the wound was closed and the animal was allowed to recover.

The opaque substances which were used were thorotrast (Testagar Company, Detroit, Michigan) and micronized barium sulfate (Micropaque) (Damaney and Company, Ware Hertz, England). Thorotrast was found to be very suitable for organs without a rich lymphatic network, such as the colon and the kidney capsule. When the lymphatic network was well developed,

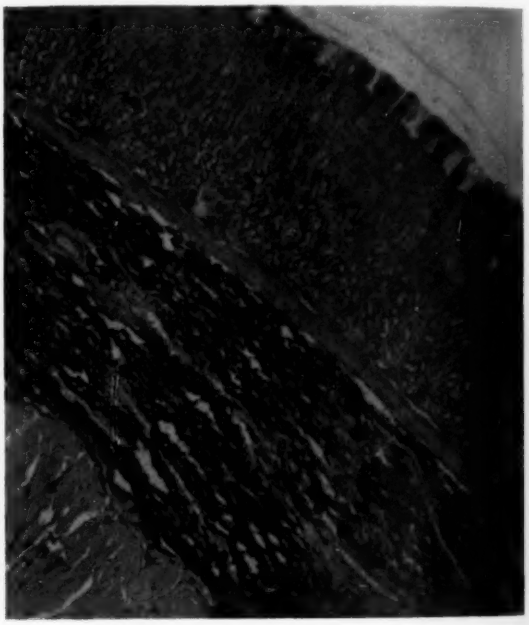

FIG. 1. Macrophages laden with thorotrast particles in the submucosa of the colon of a dog.

* Work done for U.S.A.F. contract number 29-(600)—2038.
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thorotrast was carried away before it could become fixed by the tissues. In such cases, micronized $BaSO_4$ proved to be a more satisfactory medium because it has larger particles which are not easily dispersed by the lymph vessels.

All animals prepared did well postoperatively. Roentgenograms taken soon after surgery showed a rather patchy distribution of the opaque substance, but a more even opacification was usually evident ten to twenty days later. This was especially true in the colon where the eventual appearance recalls that achieved by double contrast examinations.

Several dogs were sacrificed for gross and histologic studies at various times following surgery. All organs were grossly normal. On microscopic study, the opaque material was found to be imbedded in large macrophages. In the gastrointestinal tract most of these barium and thorotrast laden macrophages were concentrated in the submucosal layer (Fig. 1). The type of tissue reaction was essentially the same for thorotrast and $BaSO_4$.

Fourteen of the 31 animals were kept under observation for more than ten months, and all of them appeared to remain in excellent health. The illustrations show the quality of the images obtained (Fig. 2, 3, 4, and 5).

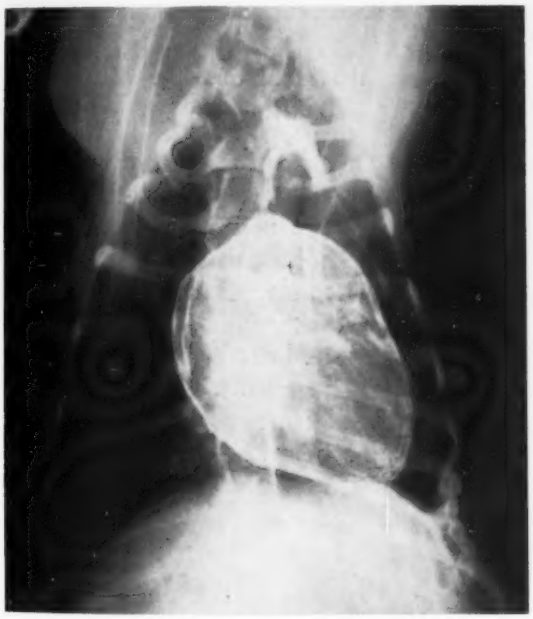

Fig. 2. Thorotrast in the pericardial wall of a dog.

Fig. 3. Thorotrast in the kidney capsules and in the wall of the left colon of a dog.

PHYSIOLOGIC APPLICATIONS

The physiologic applications of this method of opacification are manifold. In hollow muscular organs, the proper roentgenologic technique will reveal contractile patterns under normal conditions and under the influence of various stimuli, regardless of the presence and of the volume of the contents of the organs and without the misleading effects of anesthesia. In the

parenchymal organs, volume changes can be estimated from the changes in the size of their roentgen shadows. In some cases this may take the place of plethysmographic studies, which usually involve surgical exposure of the organs and general anesthesia.

CONCLUSIONS

As a general conclusion it can be stated that thorotrast and finely divided particles of $BaSO_4$ can be used in experimental animals to make permanently opacified to roentgen rays such organs as the colon, stomach, small intestine, urinary bladder, kidney, diaphragms and pericardium.

Thorotrast, a medium that is highly miscible with body fluids, has proved to be a very desirable medium for the opacification of some organs. Micropaque (finely ground $BaSO_4$) can be used as a substitute for thorotrast in other organs where the lymphatic drainage is such that thorotrast is drained away. $BaSO_4$ is not distributed in the tissues as well as thorotrast is and it gives an appearance of patchiness.

Thorotrast carries the stigma of being radioactive and therefore possibly damaging to body tissues over a period of time.[3] Histologically, the presence of thorotrast or $BaSO_4$ in the walls of an organ will produce an infiltration of macrophages, but the severity of this response is not great and does not seem to interfere with the basic physiology of the organs.

It is believed that organs permanently opacified to roentgen rays can be used for studying physiologic responses for a year or more after the material is injected.

FIG. 5. $BaSO_4$ in the stomach wall and in the wall of the bladder of a dog.

FIG. 4. $BaSO_4$ in the diaphragm of a dog.

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THE AMERICAN JOURNAL OF ROENTGENOLOGY, RADIUM THERAPY AND NUCLEAR MEDICINE

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Forty-fourth Annual Meeting: Waldorf-Astoria Hotel, New York, N. Y., April 2–4, 1962.

* *Ex officio.*

EDITORIALS

THE SIXTY-SECOND ANNUAL MEETING OF THE AMERICAN ROENTGEN RAY SOCIETY

THE Sixty-second Annual Meeting of the American Roentgen Ray Society was held at the beautiful Deauville Hotel, Miami Beach, Florida from Tuesday, September 26 through Friday, September 29, 1961.

President Harold O. Reineke issued the call to order and then introduced Dr. W. L. Fitzgerald, President of the Dade County Medical Association and Clinical Professor of Urology at the University of Miami, Florida, who delivered an Address of Welcome in which the example of scientific informational interchange embodied in this meeting was lauded. The transcendation of national boundaries and elevation of ideals above pursuit of personal aggrandizement was set forth as a model of cooperation for international diplomats and statesmen. The Society was most cordially greeted in the name of the 1,400 members of the Dade County Medical Association.

President-Elect Traian Leucutia was then installed in the office of highest honor. Dr. Ralph M. Caulk, Chairman of the Executive Council, awarded the President's Medal, and President Reineke presented two gavels; the first, a symbolic one fashioned from the tooth of a mastodon and recovered from Alaskan glacial ice, and the second, the working gavel fashioned from mementos representing the contributions of Röntgen, Coolidge and Snook to radiology

The Inaugural Address by President Leucutia titled, "Integrated Radiology," stressed the necessity of binding and recementing the major segments—diagnostic roentgenology, therapeutic radiology and nuclear medicine—into an integrated whole. Certain changes are manifest as evidenced by the development of diagnostic roentgenology into diagnostic radiology with the embodiment of such procedures in nuclear medicine. The historic expansion of the official JOURNAL of our Society amply speaks for this need and clearly documents this chain of events. As we move into the space age, further contributions to the human kind may be realized from "space-born" radiations as have been gleaned from those which are "earthborn"—both natural and artificial. Steps in progress are frequently taken by those with necessarily detailed knowledge of various Disciplines but also with a perspective of their integrated applications. President Leucutia's Inaugural Address, published elsewhere in this issue of the JOURNAL, eloquently depicts the background of the past and the implications for the future of "Integrated Radiology."

The Scientific Session of the Society began immediately thereafter with the presentation on the first morning of nine papers on Special Procedures devoted to cardiovascular roentgenology and angiographic studies. The first of these explored various aspects of the significance of protein binding of contrast media in roentgen diagnosis. Cerebral angiography, angiography in determining the resectability of lung cancer, anomalous pulmonary venous return, pericardial effusion, aortic valvular diease, cardiac ventriculography, cardiac catheterization and intravascular nonsurgical grafting, and extra large field angiography were other methods and conditions discussed.

The programming of the scientific papers in the morning provided ample time for study of the scientific and technical ex-

hibits and attendance at the Instruction Courses. The Scientific Exhibits so capably arranged by the Committee under the Chairmanship of Dr. Charles O. Heilman, Fargo, North Dakota, are described in detail elsewhere in this issue of the JOURNAL.

On Tuesday evening the Annual Caldwell Lecture was delivered by Dr. Harold O. Peterson, Chairman, Department of Radiology of the University Hospitals and Professor of Radiology, University of Minnesota, Minneapolis, Minnesota. The title of his lecture was, "The Radiologist and the Special Procedures." Dr. Peterson was introduced by Dr. J. Paul Medelman, Director, Department of Radiology, St. Joseph's Hospital, St. Paul, Minnesota, and Clinical Associate Professor of Radiology, University of Minnesota, Minneapolis, Minnesota. Dr. Peterson's Lecture described the many facets of special diagnostic procedures performed by the radiologist and he emphasized that these procedures can often best be executed by the radiologist. The exceptionally large attendance and enthusiasm with which the audience received this Forty-first Annual Caldwell Lecture obviously expressed the importance of these procedures in present day diagnostic roentgenology and was a fine tribute to the outstanding abilities of Dr. Peterson. At the conclusion of the Lecture, President Leucutia presented to Dr. Peterson the Caldwell Medal, the highest award that our Society can offer for meritorious scientific achievement.

The Scientific Session was resumed on Wednesday morning with Dr. Ralph M. Caulk presiding. Most of the papers of this Session dealt with various aspects of diagnostic roentgenology with the last being a discussion of radiation hazards. It was emphasized that earlier predictions of biologic damage from radiation sources were unduly pessimistic and the radiologist was charged with the responsibility of realistically interpreting these various hazards.

The Scientific Session on Thursday morning with Dr. Clyde A. Stevenson presiding was also on various aspects of diagnostic roentgenology. One of the most interesting described the examination of the lower human colon by the use of a silicone foam. A soft mold is formed after its instillation which upon expulsion reveals imprints of mucosal abnormalities. In some instances cytologic examination can be carried out by recovering cells from the surface of the mold.

On Thursday afternoon a special instruction course, in the form of a Panel Discussion, on "Current Opinions from the American College of Radiology on Principles of Ethics, Economics and Facts and Figures" attracted a surprisingly large audience. Dr. Earl E. Barth moderated the Panel consisting of Drs. Bailey, Buchanan, Present and Wachowski. Dr. L. H. Garland, President of the American College of Radiology also participated in the timely discussion of these extremely important matters pertaining to radiologic practice which are of particular value to us younger radiologists.

On Thursday evening the Annual Banquet was held in the Casanova Room in a festive Caribbean atmosphere created by an excellent floor show of singing and interpretive dancing. The newly elected officers were announced with Dr. Earl E. Barth being the President-Elect of the Society. Awards were presented for those scientific exhibits judged to be outstanding.

On Friday morning with Dr. Robert C. Pendergrass presiding, ten papers on radiation therapy, one on the use of P^{32} in the evaluation of ocular neoplasms and one on newer trends in radiation therapy completed the program of the Scientific Sessions.

The Director of Instruction Courses, Dr. Harold O. Peterson, assisted by Dr. Donn G. Mosser, Associate Director, assembled an excellent array of courses, many of which dealt with the special procedures which Dr. Peterson discussed in the Caldwell Lecture. The fact that most were "sold-out" testifies to their popularity and value.

The number of technical exhibits was the

largest in the Society's history with complete utilization of the available space. These were assembled by Mr. Clifford L. Sherratt, Honorary Member, and Dr. James C. Cook, Manager of the Annual Meeting. A detailed list of the Technical Exhibits is published elsewhere in this issue of the JOURNAL.

The locale of the meeting in such a famous resort city as Miami Beach, along with the highly efficient and able efforts of the Committee on Local Arrangements, afforded delightful diversions for the ladies attending the meeting. On Monday, a Golf Widows' Boat Cruise to Fort Lauderdale and a tour of the tropical surroundings at Patricia Murphy's Candlelit Garden was capped by an excellent dinner. On Tuesday a tour to the Parrot Jungle with its exotically beautiful flowers and birds and a visit to the Seaquarium was coupled by lunch at the Key Biscayne Hotel. Wednesday found the ladies again cruising, this time toward the Villa Viscaya (an "Italian Palace in Miami") with its incomparable tapestries and furnishings, tropical gardens and statuary, many of which were imported from various castles in Europe, particularly Italy. The unusually good attendance on all of these trips was an obvious expression of the Committee's success. The Society is deeply appreciative of the diligent efforts of Mrs. Gerard Raap, Chairman, and Mrs. John S. Stewart, Co-Chairman and their charming Ladies Committee on Local Arrangements.

The Annual Golf Tournament, held at the La Gorce Country Club on Monday afternoon, was arranged by the Local Golf Committee under the chairmanship of Dr. Virgil H. Pieck. This course, one of the most scenic and beautiful on which this tournament has ever been held, afforded an enjoyable afternoon followed by the Golf Dinner at which the prizes were awarded.

To the Management and Staff of the Deauville Hotel the Society extends its sincere thanks for excellent service, a most pleasant atmosphere and the splendid manner in which all details of the meeting were carried out.

The Committee on Press Relations with Dr. David Kirsh as Chairman, and assisted by Mr. Otha W. Linton and Mr. David Pettengill from the American College of Radiology, ably handled press releases and public relations.

Dr. Gerard Raap deserves special commendation for his enthusiastic Chairmanship of the Committee on Local Arrangements. He truly embodied southern hospitality in its finest sense.

President Leucutia can feel pardonably proud for an eminently successful meeting which will be long remembered.

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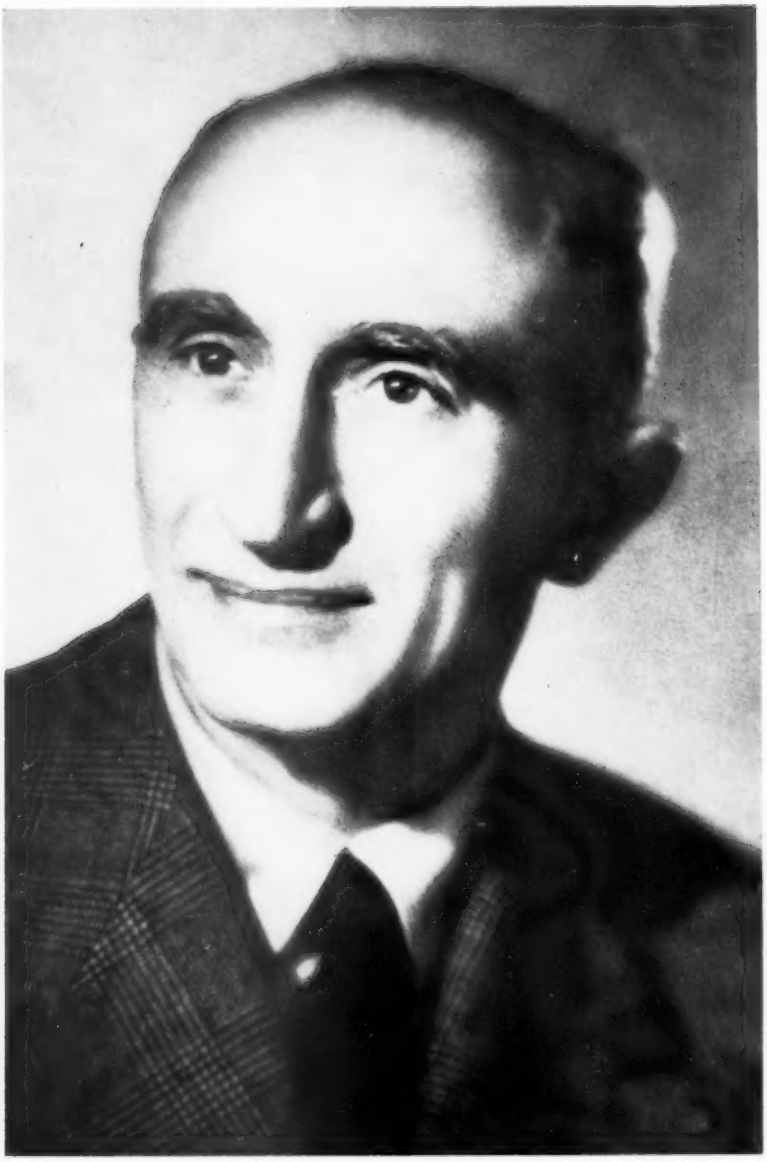

G. G. PALMIERI
1892-1961

PROFESSOR PALMIERI, who was born on February 6, 1892 in Bologna, Italy, received his medical degree in 1916 from the University of the same city. He was named Director of the Department of Medical Radiology of the University of Bologna in 1924, first in the capacity of Associated Professor and since 1939 as a full time Professor. In 1930 he also became Director of the L. Galvani Radium Institute of Bologna.

Professor Palmieri was Vice-President of the Società Italiana di Cancerologia in 1957 and of the Società Italiana di Radiobiologia

Medica in 1959. His many honors included the title of Accademico Benedettino dell'Accademia delle Scienze dell'Istituto di Bologna; honorary member of the Caroline-Leopold Academy of Halle and of the Medical Academy of the Republic of Colombia; honorary member of the Radiologic Societies of Germany, Austria, Czechoslovakia, Hungary, Spain, Mexico and Colombia; corresponding member of the Accademia di Scienze, Lettere e Arti di Modena and member of the Honorary Committee of Centre Antoine Béclère of Paris. He was an ardent supporter of the International Congress of Radiology and on two occasions headed the Italian delegation; in 1959 he was awarded the coveted Roentgenplakette in Monaco.

Professor Palmieri published more than 400 scientific papers pertaining to diagnostic and therapeutic radiology and was keenly interested in radiobiology. He gave freely of his time to lectures on many facets of practical radiology, took an enthusiastic part in scientific discussions, and was a devoted teacher with a gift of communicating thoughts of inspirational knowledge along with ideas of technical proficiency.

With the death of Professor Palmieri on August 16, 1961, the Italian Radiologic Society as well as world Radiology loses a brilliant scientist, an altruistic and affectionate physician, and a truly great man.

Giulio Tori, M.D.

We regret to announce the death on September 4, 1961, at Baltimore, Maryland, of Dr. John W. Pierson, distinguished member and Past President of the American Roentgen Ray Society.

THE SCIENTIFIC EXHIBITS

THE SCIENTIFIC EXHIBITS of the Sixty-second Annual Meeting of the American Roentgen Ray Society held at the Deauville Hotel, Miami Beach, Florida, September 26–29, 1961, were of extremely high quality and most instructive. This year there was a considerable increase in the self-contained exhibits and this, with the beautiful decor of the Peacock Alley where they were displayed, immeasurably enhanced their attractiveness. The subjects of unusual technical achievements which were presented—many reflecting the importance of special procedures which were so vividly depicted by the Caldwell Lecturer of this meeting—furnished incontrovertible evidence of the extraordinarily rapid progress made in the science of radiology during the last few years. The Committee on Scientific Exhibits, under the very able chairmanship of Dr. Charles O. Heilman, Fargo, North Dakota, which spared no time or effort in making all the arrangements, has been amply compensated by the great interest shown by the members of the Society and their guests in these as well as in many other subjects of like documentary caliber. The Committee on Awards must have had a difficult task in selecting the exhibits most deserving of special merit. After careful and thoughtful consideration, one Silver Medal, one Bronze Medal, and eight Certificates of Merit were awarded.

Silver Medal

Lymphography; Lymphangio-Adenography. Manuel Viamonte, Jr., M.D., Manuel Soto, M.D., and Raymond E. Parks, M.D., Jackson Memorial Hospital and University of Miami School of Medicine, Miami, Florida.

The technique of the roentgenographic visualization of the lymph vessels and lymph nodes was illustrated in color transparencies. Two whole body roentgenograms and selected roentgenograms of conventional size illustrated the normal and abnormal findings in the clinical application of this technique.

The method is of definite value because: (1) it contributes to the detection and evaluation of the extension of lymph node metastasis; (2) it aids in the differential diagnosis of intra-abdominal and pelvic masses; (3) it assists in the evaluation of surgical, chemotherapeutic and radiotherapeutic management of malignant neoplasms; (4) it provides information as to the cause of obstructive edema, especially when combined with phlebography; and (5) it facilitates the diagnosis and treatment control of lymphomas.

The opacification of the lymph nodes is shown to better advantage on the 24 to 48 hour roentgenograms. The nodes remain radiopaque 6 to 8 weeks and in some cases as long as 6 months.

Delayed healing and infection of the wound are occasionally seen. Also, a miliary pattern may be observed in the lungs several hours after the injection, due to interstitial deposition of the ethiodol which is used as the opaque medium but this was found to be of no clinical significance.

It was concluded that lymphography represents a dynamic approach to the study of the anatomy, physiology and pathology of the lymphatic system with vast clinical and experimental potentialities.

Bronze Medal

Cerebral Angiography in Medical Practice; A Compendium of Clinical and Neuroroentgenologic Features. C. B. Holman, M.D., H. L. Baker, Jr., M.D., A. Uihlein, M.D., R. H. Miller, M.D., and A. H. Bulbulian, D.D.S., Mayo Clinic and Mayo Foundation, Rochester, Minnesota.

In this exhibit the angiographic patterns of most of the intracranial tumefactive and vascular lesions were presented. The diagnostic features on the angiograms were readily apparent, because each was accompanied by a simplified explanatory diagram.

Information relating to the choice of patients, injection procedures and clinical complications was given *in extenso*.

CERTIFICATES OF MERIT

1. *The Detection of Upper Gastrointestinal Tract Pathology with High Kilovoltage Techniques*. Steven J. Figiel, M.D., Leo S. Figiel, M.D., Fred K. Wietersen, M.D., E. Lievois, M.D., D. K. Rush, M.D., and R. M. Balow, M.D., The Grace Hospital, Detroit, Michigan.

High kilovoltage techniques and standard techniques were compared by presenting numerous cases. Roentgenographic studies of the surgical material with varying kilovoltages and Kodachromes of the surgical specimens were used to establish a final correlation between the roentgenologic and pathologic picture.

2. *Skeletal Abnormalities Associated with Gonadal Dysgenesis*. Nathaniel Finby, M.D., and Reginald MacGregor Archibald, M.D., Rockefeller Institute, New York, New York.

Gonadal dysgenesis is a genetic syndrome usually associated with an absent sex chromosome and a resultant XO pattern. Few of these patients have the classical "Turner's syndrome" but most do show skeletal abnormalities which are helpful in diagnosis. The abnormalities include dwarfism, slight retardation of skeletal maturation, metacarpal or carpal abnormalities with a positive "metacarpal sign," cubitus valgus and a "tibia-vara-like" deformity of the knees. The very young patient may show soft tissue edema and the older patient may show "osteoporosis."

The exhibit consisted of illustrative cases demonstrating these skeletal abnormalities.

3. *Juvenile Rheumatoid Arthritis*. William Martel, M.D., John F. Holt, M.D., and James T. Cassidy, M.D., University of Michigan, Ann Arbor, Michigan.

Certain roentgen manifestations of juvenile rheumatoid arthritis which differ from those observed in the adult type of the disease formed the theme of this exhibit.

Growth disturbances may be severe and periosteal new bone formation is frequent. Fractures and subluxations are often present. The atypical distribution of involved joints and the tendency toward cervical ankylosis are significant.

Other conditions requiring consideration in the differential diagnosis were also discussed.

4. *Bronchography: Diagnosis of Localized Peripheral Lesions*. William Molnar, M.D., Atis K. Freimanis, M.D., and William M. Blackwell, M.D., Ohio State University Hospital, Columbus, Ohio.

A method using an aerosol nebulizer for the introduction of topical anesthesia was described. The value of bronchography in the differential diagnosis of localized peripheral pulmonary lesions, not accessible to bronchoscopy, was illustrated by representative findings on bronchograms selected from a series of some 200 problem cases. The relatively greater extent of the bronchial divisions "seen" by bronchography as compared to bronchoscopy was emphasized.

5. *An In Vivo Dosimetry System for Clinical and Experimental Radiation Therapy*. Bernard Roswit, M.D., Stanley J. Malsky, M.A., M.Sc., Charles Amato, B.Sc., Cyprian B. Reid, B.Sc., Charles Spreckels, B.Sc., and Louis Maddalone, Mold Technologist, Veterans Administration Hospital, Bronx, New York.

The authors have designed and employed an *in vivo* human dosimetry system which has proved to be accurate and dependable and is relatively inexpensive. This system includes a detector, a reader, and a variety of tubes, needles, catheters and molds for *in vivo* applications.

The procedure was used in nearly every anatomic organ, tissue and space during clinical and experimental radiation therapy.

This exhibit described in detail the experience gained with the physical development and clinical use of the system.

6. *Tumors of the Small Intestine*. W. G. Sauer, M.D., W. H. Dearing, M.D., C. G. Moertel, M.D., M. B. Dockerty, M.D., C. A. Good, M.D., and A. H. Bulbulian,

D.D.S., Mayo Clinic and Mayo Foundation, Rochester, Minnesota.

Tumors of the small intestine are relatively uncommon and often present a diagnostic enigma to the physician. The authors have reviewed more than 700 tumors of the small intestine encountered at the Mayo Clinic in the 20 year period ending January 1, 1958. These tumors have been classified according to benignancy or malignancy, site or origin and relative frequency.

The commonest benign tumors in this group include leiomyoma, adenoma, lipoma and hemangioma; the commonest malignant neoplasms are carcinoid tumors, adenocarcinoma, malignant lymphoma and leiomyosarcoma.

This exhibit detailed the clinical features, laboratory and roentgenologic findings, gross and histologic characteristics, and the treatment recommended in each of these eight types of neoplasm. It also briefly outlined lesions that are seen much less frequently.

7. *Clinical Applications of Lymphangiography.* Sidney Wallace, M.D., Laird Jackson, M.D., Burton Schaffer, M.D., R. John Gould, M.D., Roy R. Greening, M.D., Simon Kramer, M.D., and Arthur Weiss, M.D., Jefferson Medical College Hospital, Philadelphia General Hospital, Philadelphia, Pennsylvania.

Lymphangiography by direct intralymphatic injection depicts the lymph vessels and lymph nodes of the extremities, inguinal, external and common iliac, and para-aortic areas, as well as of the axillary and supraclavicular areas and the thoracic duct.

In this exhibit the most useful clinical applications were demonstrated in detail.

8. *Fluid-Filled Loops in Intestinal Obstruction.* John L. Williams, M.D., and George A. Moller, M.D., The George F. Geisinger Memorial Hospital, The Foss Clinic, Danville, Pennsylvania.

In 6 per cent of 300 patients with intestinal obstruction, loops were filled with fluid but little or no gas. The incidence of gangrene is quite high in these fluid-filled obstructions and, therefore, the most dangerous cases are often the most difficult to diagnose. The correct roentgen diagnosis depends upon the recognition of the abnormal soft tissue densities caused by these fluid-filled loops. The abdomen is usually not distended and has no auscultatory signs.

The exhibit presented some unusual cases illustrating these points.

Other Exhibits

Other excellent exhibits were:

Radiology on Stamps. Stanley P. Bohrer, M.D., Massachusetts General Hospital, Boston, Massachusetts.

This exhibit consisted of first day covers, plate blocks, and other philatelic *objets d'art* documenting radiologic, nuclear, and atomic historic events. Correspondence from interested medical philatelists would be welcomed by the exhibitor.

The Value of the Admission Chest Film As an Aid in the Diagnosis of Extracardiac and Extrapulmonary Diseases in Children. Donald H. Altman, M.D., and Robert F. Conti, M.D., Variety Children's Hospital, Miami, Florida.

A group of 18 pediatric cases was discussed in which the admitting chest roentgenograms could be used to make a specific diagnosis, or strongly suggest the diagnosis, of extracardiac and extrapulmonary disease, later to be confirmed by additional studies.

The careful evaluation of all information available on the chest roentgenogram can frequently be a clue to obscure diseases in childhood even though the clinical information may be scant.

Dysgenesis of the Proximal Femur: Congenital Short Femur, Bowed Femur, and Coxa Vara. Harlan C. Amstutz, M.D., and Philip D. Wilson, Jr., M.D., Hospital for Special Surgery, Cornell Medical Center, New York, New York.

The development and variability of femoral neck-shaft valgus and version were presented. The roentgen characteristics of

coxa vara as a result of many acquired and congenital disturbances were illustrated. The roentgen features, pathogenesis, prognosis and treatment of three groups of seemingly related congenital ossification disturbances (congenital short femur with coxa vara, congenital bowed femur with coxa vara, and congenital coxa vara) were emphasized.

Azygography: Its Value in Mediastinal Metastases and Tumors. Arnold L. Bachman, M.D., Wolfgang Ackermann, M.D., and Kevin L. Macken, M.D., Columbia University, College of Physicians and Surgeons, New York, Yew York.

Roentgenographic demonstration of mediastinal metastases by the visualization of alterations and obstructions in the azygos venous system has proved to be of great importance in the determination of operability in carcinoma of the lung and esophagus.

In this exhibit were included the method of performing azygography, a description of the normal appearance and variations of the azygos veins, their appearance in nonneoplastic disease, the findings in carcinoma of the lung and esophagus, and in mediastinal tumors. Correlation with surgical and autopsy data was made.

The Mastoid. Joshua A. Becker, M.D., and Henry J. Woloshin, M.D., Temple University Medical Center, Philadelphia, Pennsylvania.

The resurgence of middle ear and mastoid surgery has brought new emphasis on mastoid roentgenography.

The authors attempted to establish a diagnostic regimen that will give the maximum amount of information, but which can be done with conventional roentgenographic equipment and without undue complexity. They obtained good preoperative correlation in 58 per cent of the cases with the surgical pathology findings. Further review yielded a 72 per cent correlation.

The exhibit gave a detailed account of the results.

The Post-Mortem Coronarogram. Donn J. Brascho, Captain, M.C., USA, Brooke General Hospital, Brooke Army Medical Center, Fort Sam Houston, Texas.

The purpose of this exhibit was to demonstrate the results of a new and simple method for postmortem roentgenographic evaluation of the arterial supply of the human heart.

Improved Results in Treatment of Oral Carcinoma through Use of Precision Therapy with Custom Made Cones. S. Gordon Castigliano, M.D., and C. Jules Rominger, M.D., American Oncologic Hospital and Misericordia Hospital, Philadelphia, Pennsylvania.

The exhibitors presented improved results in the treatment of oral carcinoma during the last seventeen years using precision therapy with custom-made cones.

Protection can be assured to important uninvolved structures and adequate dosage can safely be delivered to well controlled volumes. Precision protection of adjacent tissues allows the performance of surgical procedures later with perfect healing.

Radioactive Phosphorus (P^{32}) for Diffuse Metastatic Bone Disease: The Third Decade of Use. Harold Chiat, M.D., and Perry Mandel, M.D., Nassau Hospital, Mineola, New York.

The technique of combined radioactive phosphorus (P^{32}) and testosterone for diffuse metastatic bone disease has had sufficient clinical evaluation to establish it as an important and rewarding therapeutic modality.

Representative cases were included in this exhibit with emphasis on the proper selection of patients, the technique of administration, the hazards, and the results.

Recent studies have clarified the involved physiologic mechanisms which suggest new areas worthy of further investigation.

Simple, Safe and Reliable Radiography of Infants. Donald B. Darling, M.D., Boston City Hospital, Boston, Massachusetts.

A method of obtaining the most routine studies in infants without radiation exposure to personnel was illustrated. The average gonad dose as measured during actual

exposure was indicated in each case. High speed screens, high speed films, 3 mm. Al filtration, double diaphragm collimation with full field illumination and 1/16 inch lead gonadal shields were employed.

The Solitary Dense Vertebral Body. John M. Dennis, M.D., University of Maryland, School of Medicine, Baltimore, Maryland.

Hodgkin's disease, Paget's disease and metastatic malignancy occasionally produce a solitary dense vertebral body. The etiology of this dense vertebral body can usually be determined roentgenographically from the bony architecture. These architectural changes produced by the three diseases were demonstrated.

Combined Roentgenologic and Gastroscopic Evaluation of Gastric Ulceration. Gerald D. Dodd, M.D., and Robert S. Nelson, M.D., University of Texas, M. D. Anderson Hospital and Tumor Institute, Houston, Texas.

Combined roentgenologic and gastroscopic evaluation of gastric ulcers has proved highly accurate. A correlation of the significant differential points of each method of examination was illustrated by roentgenograms and color photographs made through the gastroscope. A statistical analysis of a series of 100 cases was presented.

Cranial Nerve Metastases in Tumors of the Head and Neck. Gerald D. Dodd, M.D., and Patrick A. Dolan, M.D., University of Texas, M. D. Anderson Hospital and Tumor Institute, Houston, Texas.

Perineural and endoneural lymphatic vessels have proved to be important but neglected routes of spread in tumors of the head and neck. Existing therapeutic procedures can be modified to meet this contingency provided the clinician is aware of their actual or potential presence.

In approximately one-third of 37 proved cases at the M. D. Anderson Hospital, pretreatment recognition has proved feasible by plain film examination of the skull and facial bones by changes in the various transmittal canals and foramina of the major divisions of the cranial nerves. The roentgen anatomy of the most frequently involved nerves and the changes encountered with neural metastases were detailed.

Ectopic Ureterocele (Ectopic Ureter with Ureterocele). John P. Dorst, M.D., and Frederic N. Silverman, M.D., Departments of Radiology and Pediatrics, University of Cincinnati College of Medicine, Cincinnati, Ohio.

The most common type of ureterocele found in infants and children is an intravesical protrusion of the anterior aspect of a dilated ureter which passes between the mucosa and muscular coat of the bladder before it ends in the urethra. Termed an ectopic ureterocele, it differs from the better known simple ureterocele which characteristically occurs in the adult, in pathologic anatomy, size, severity of symptoms, prognosis, and therapy.

The pathologic anatomy and roentgenographic appearance of the two forms of ureterocele were contrasted, and the most common pitfalls in the diagnosis of the ectopic ureterocele were illustrated.

Flow Guided Catheterization and Nonsurgical Grafting: Two New Techniques for Cardiovascular Diagnosis and Treatment. Charles T. Dotter, M.D., and Kurt R. Straube, M.D., University of Oregon Medical School, Portland, Oregon.

The use of flexible tubing in place of ordinary cardiac catheters provides a simple means for downstream or flow guided catheterization of the pulmonary systemic arteries. The determination of man's pulmonary artery pressure requires neither skill nor fluoroscopy and can be done as a ward or office procedure.

Percutaneous spring guided arterial catheterization affords simple, experimentally established means for the nonsurgical insertion of silastic sleeve grafts into coronary, carotid, renal and peripheral arteries. If our "pipe dreams" are realized, this approach may assume considerable significance in the practice of medicine.

The exhibit demonstrated the two techniques.

An Evaluation of Roentgen Signs of Pan-

creatic Enlargement. William R. Eyler, M.D., Max D. Clark, M.D., and Roger L. Rian, M.D., Henry Ford Hospital, Detroit, Michigan.

This exhibit consisted of selected roentgenograms from over 100 cases of pancreatic enlargement (*i.e.*, carcinoma, pancreatitis, pseudocysts and other less common causes) demonstrating the various roentgen signs. There was also a brief summary of some of the associated clinical findings.

The Colonic Polyp and Colonic Cancer. Detection, Significance and Roentgenographic Follow-Up of Colonic Polyps with the Use of High Kilovoltage Compression Technique. Steven J. Figiel, M.D., Leo S. Figiel, M.D., Fred K. Wietersen, M.D., Thomas Arminski, M.D., and D. W. McLean, M.D., The Grace Hospital, Detroit, Michigan.

The spot compression high kilovoltage method for detection of the colonic polyp was used to illustrate the development of colonic cancer from the colonic polypoid lesion.

Pertinent autopsy and histopathologic data were included. The authors presented comprehensive case material demonstrating their approach in determining the necessity for surgery when one or more colonic polyps are present.

Closed Circuit Television Tape and Direct Kinescopic Recording of the Fluoroscopic Image. Bertram R. Girdany, M.D., and Edwin S. Gaither, M.D., Children's Hospital of Pittsburgh, Pittsburgh, Pennsylvania.

The individual components that make up a closed circuit television fluoroscopic system and tape recorder were explained in words and diagrams. The advantages of television tape recording were listed. Table top radiation dosages during actual recording were tabulated. A live kinescopic film was shown.

The Pineal Laminagram: Visualization of the Faintly Calcified Pineal Gland. John A. Goree, M.D., Karl K. Wallace, Jr., M.D., and Richard L. Bean, M.D., Department of Radiology, Duke University Medical Center, Durham, North Carolina.

The small pineal calcification is usually clearly visible on a lateral roentgenogram of the skull but is often obscured in all anteroposterior and posteroanterior projections, including stereoscopic films. Laminagraphy will demonstrate pineal calcification in many such instances and make localization of this structure possible. A simple technique and illustrative cases were presented.

Roentgen Findings in Diseases with Abnormal Calcifications. Theodore F. Hilbish, M.D., and Frederic C. Bartter, M.D., National Institute of Health, Clinical Center, Bethesda, Maryland.

This exhibit presented the roentgen findings of diseases associated with abnormal calcium deposition including such conditions as metastatic calcification, calcinosis, dystrophic calcification, heterotopic bone formation, vascular calcification and stone formation. The exhibit was intended to portray the interesting and, at times, unusual calcium depositions observed within the body tissues. Brief comments concerning the diagnosis were included in the exhibit.

Intercalated Segmental Pulmonary Arteriography. J. C. Hirschman, M.D., Manuel Viamonte, Jr., M.D., R. J. Boucek, M.D., and Raymond E. Parks, M.D., Jackson Memorial Hospital and University of Miami School of Medicine, Miami, Florida.

This method of angiography, which makes use of electronically programmed injection of the opaque medium and the roentgen ray exposure, was applied to the study of pulmonary vascular physiology and pathology. Experimental work in dogs and humans has demonstrated the presence of pulmonary vasomotor responses and some more permanent pathologic changes in various disease states.

Venography in Thrombophlebitis of the Lower Extremity: A Useful Techniuqe in Diagnosis and in the Evaluation of Thrombolytic Agents. Richard A. Kramer, M.D., Sydney P. Hecker, M.D., and John F.

Weigen, M.D., Palo Alto Medical Clinic (Departments of Radiology and Internal Medicine), Palo Alto, California.

Venography of the lower extremity is useful both in establishing the diagnosis of thrombophlebitis and in evaluating the efficacy of treatment. Many conditions can simulate the clinical appearance of thrombophlebitis, and in equivocal cases venography can be a valuable diagnostic adjunct to the usual clinical studies. Venography also offers an objective way of comparing the efficacy of various thrombolytic agents.

Plasmin is currently being evaluated in the treatment of stroke, coronary occlusion and peripheral arterial occlusion in addition to thrombophlebitis. Angiographic techniques will be invaluable in these studies.

This exhibit illustrated valuable points pertaining to these studies.

Polydirectional Body-Section Radiography with the Polytome. J. T. Littleton, M.D., C. L. Rumbaugh, M.D., and F. S. Winter, M.D., The Guthrie Clinic and Robert Packer Hospital, Sayre, Pennsylvania.

This exhibit served to introduce the use of polydirectional body-section roentgenography in all of its phases through the use of the Polytome. This is the first unit of its type to be employed in the United States, although it has become a fairly common method in Europe. It is the feeling of the authors that this instrument offers a diagnostic advantage which has heretofore not been fully appreciated in the United States.

Contrast Roentgenography of the Larynx. Jose T. Medina, M.D., William B. Seaman, M.D., Ira Kanter, M.D., P. Carbajal, M.D., and D. Baker, M.D., Columbia-Presbyterian Medical Center, New York, New York.

The technique of the roentgen examination of the larynx and hypopharynx by the instillation of oily dionosil after local anesthesia was described. The resultant coating of the mucosa produces a double contrast image of the larynx, hypopharynx and upper trachea with anteroposterior and lateral films exposed (1) during quiet breathing through the nose with the mouth closed, (2) during phonation of the vowel Eeeee, (3) while performing the true Valsalva maneuver, and (4) while performing a modified Valsalva maneuver by blowing through the closed lips.

Examples of the normal anatomy, anatomic variants, and various pathologic states have been selected for exhibition from a total clinical materal of almost 200 patients.

Intravenous Abdominal Arteriography. William T. Meszaros, M.D., John B. Graham, M.D., Howard C. Burkhead, M.D., Myles P. Cunningham, M.D., George C. Sutton, M.D., and Brown Brooks, M.D., Cook County Hospital, Evanston Hospital, and Northwestern University Medical School, Chicago and Evanston, Illinois.

Intravenous arteriography is a relatively new method of visualizing the abdominal aorta and its branches. Roentgenologic examination of the aorta is performed following intravenous injection of contrast material. The value and limitations of this procedure were shown on films illustrating various types of vascular abnormalities.

Detection of Regional Lung Function and Early Carcinoma by Fluorodensimetry. Charles P. Oderr, M.D., Veterans Administration Hospital, New Orleans, Louisiana.

Simultaneous electronic recording of fluoroscopic density during breathing maneuvers in two similar areas makes it possible to detect a relative impairment of function in one. The record made reveals the change in density with quiet, deep and fast breathing as well as during cough. The loss of amplitude in expansion and contraction of a given lobe is probably earlier evidence of endobronchial disease, such as carcinoma, than either atelectasis or emphysema.

These pertinent points were demonstrated in the exhibit.

The Problem of the Overexposed Film: Practical Methods of Film Reduction. An-

drew K. Poznanski, M.D., C.M., Henry Ford Hospital, Detroit, Michigan.

Many of the well known photographic reducers are not ideally suited for the relatively high density, high contrast, double emulsion roentgen-ray film. A search was made for a reducer that would be effective on roentgen-ray film and be simple to use. A copper sulphate reducer was found to be simple to use, stable when stored, not highly toxic and its components are readily available.

In this exhibit the procedure was described in detail and other methods of film reduction, including removal of one emulsion and the use of the logEtron, were compared.

Parosteal Osteoidsarcoma. Klaus Ranniger, M.D., Department of Radiology, University of Chicago, Chicago, Illinois.

The roentgenologic appearance and the clinical behavior of parosteal osteoidsarcoma (juxtacortical osteosarcoma) are distinctly different from other bone sarcomas, particularly from the usual type of osteosarcoma. Eleven cases with follow-up examinations were presented which demonstrated the typical roentgenologic features and the slow progression of this variant of osteosarcoma.

Shelf Tumors and Their Differential Diagnosis. Julian O. Salik, M.D., Sinai Hospital of Baltimore, Inc., Baltimore, Maryland.

Pathologic changes in the recto-uterine or rectovesical excavation may be caused by benign or malignant tumors or may be of inflammatory origin. The roentgen appearance and differential diagnosis in male and female were illustrated by representative cases.

Case of the Day. Stefan C. Schatzki, M.D., Walter Reed General Hospital, Washington, D. C.

A "Case of the Day" for interpretation by those attending the meeting was presented. On the subsequent day the pathologic findings were given and discussion of the previous day's films was held.

This exhibit brought attention to a potential method of teaching that has proved successful in several Departments of Radiology.

Lesions Presenting in Right Cardiophrenic Angle. Hildegarde Schorsch, M.D., Edward A. Piszczek, M.D., M.P.H., and Ki Eun Chung, M.D., Cook County Hospital, Stritch School of Medicine, Loyola University, The Suburban Tuberculosis Sanitarium District, Chicago, Illinois.

The differential diagnosis of lesions which project into the right cardiophrenic angle in the plain chest roentgenogram was discussed.

These lesions, based on etiology, were classified into 7 groups: (1) upper gastrointestinal; (2) diaphragmatic hernia; (3) pulmonary; (4) cardiovascular; (5) pleural; (6) intrathoracic tumors; and (7) miscellaneous.

Representative cases were illustrated.

Estimation of Experimental Coronary Hemodynamics by Coronary Cine-Arteriography. William H. Sewell, M.D., Veterans Administration Hospital, Oteen, North Carolina.

Coronary cinearteriograms were taken at 30 frames per second on 35 mm. Plus-X film by injecting 90 per cent hypaque into the sinus of Valsalva of dogs. Both the diameter and rate of movement of the medium in the anterior descending artery were compared with the circumflex, before and after various degrees of surgical construction of the anterior descending branch. After the standard deviation of the method was determined, it was possible to tell whether a surgical constriction had significantly reduced the relative flow in the anterior descending artery and to estimate the degree of change.

The exhibit demonstrated the results obtained in detail.

The Differential Diagnosis of Vertebral Body Compression. William C. Strittmatter, M.D., and Barry Q. Walker, M.D., Cleveland Clinic Foundation, Cleveland, Ohio.

In this exhibit some of the lesions that cause compression of vertebral bodies were

presented. They were divided into six main groups (1) traumatic, (2) infectious, (3) neoplastic, (4) metabolic, (5) vascular; and (6) systemic disorders of unknown etiology.

Representative examples of each group were included in an attempt to aid the radiologist in arriving at a correct diagnosis.

Roentgenographic Examination of the Vocal Cords. Sol M. Unger, M.D., Harold G. Jacobson, M.D., George A. Meyer, M.D., H. Kolson, M.D., and Maxwell H. Poppel, M.D., Veterans Administration Hospital, Bronx, New York.

Roentgenographic examination of the larynx can easily be carried out by means of laminagraphy, or by conventional spot films during fluoroscopy. Vocal cord paralysis secondary to recurrent laryngeal nerve paralysis and vocal cord irregularity due to carcinoma of the larynx can be differentiated and this was demonstrated by the exhibitors.

The normal and abnormal larynx can also be adequately visualized on conventional roentgenograms. The anatomy of the larynx was thoroughly and clearly discussed.

A Second Look Is Worth 1,000 X-Rays: Tuberculosis Program. U. S. Department of Health, Education, and Welfare, Public Health Service, Washington, D. C.

The case for dual reading of roentgenograms in tuberculosis case finding was presented. Facilities were available at the exhibit to allow physicians to check their interpretations of sample films against those of experts. In an accompanying pamphlet studies were cited showing that dual reading increases the yield by 10 per cent or more thereby reducing radiation exposure.

The Roentgenographic Determination of Pulmonary Hypertension in Mitral Stenosis. Manuel Viamonte, Jr., M.D., Frank Barreras, M.D., and Raymond E. Parks, M.D., Jackson Memorial Hospital and University of Miami School of Medicine, Miami, Florida.

In this exhibit the criteria used for predicting the mean pulmonary artery pressure in patients with mitral stenosis were presented. Examples of cases with absent, minimal, moderate and severe pulmonary arterial hypertension were shown. Other pulmonary findings in mitral stenosis were also demonstrated. Postoperative studies should be performed 6 months to 1 year after surgical intervention.

THE TECHNICAL EXHIBITS

The Technical Exhibits of the Sixty-second Annual Meeting of the American Roentgen Ray Society, held in Miami Beach, Florida, September 26–29, 1961 were the largest and most attractive in the Society's history. The matchless convention engineering concept embodied in the Deauville Hotel, the spacious exhibit halls, Napoleon Room No. 1 and 2, and their location in the immediate vicinity of the activities of the Scientific Assembly were pleasant features equally welcomed by the Exhibitors and the many members and guests of the Society who visited them. The displays, which were artistically arranged and unprecedently beautiful, evoked the greatest admiration and thereby contributed notably to the over-all success of the meeting.

The untiring efforts of Mr. Clifford L. Sherratt, Honorary Member, and Dr. James C. Cook, Manager of the Annual Meeting were again amply rewarded by the fact that the ultramodern facilities of the Deauville Hotel were utilized to fullest extent. The Society extends its most sincere thanks to the many exhibitors for their participation at this Sixty-second Annual Meeting and regrets that, despite the increased space, a few of the applicants could not be accommodated.

The following are the firms which took part in the Technical Exhibits:

Abbott Laboratories, North Chicago,

Illinois; Ansco Division, General Aniline & Film Corporation, Binghamton, New York; Atomic Energy of Canada Limited, Ottawa, Canada; Automatic Seriograph Corporation, College Park, Maryland; Barnes-Hind Barium Products Company, Sunnyvale, California; Bar-Ray Products, Inc., Brooklyn, New York; Bell-Craig, Inc., New York, New York; Carr Corporation, Santa Monica, California; Cordis Corporation, Miami, Florida; Lester A. Dine Company, Woodside, New York; Dunlee Corporation, Bellwood, Illinois; E. I. du Pont de Nemours & Company, Inc., Wilmington, Delaware; Eastman Kodak Company, Rochester, New York; Encyclopaedia Britannica, Miami, Florida; Eureka X-Ray Tube Corporation, Chicago, Illinois; Oscar Fisher Company, Inc., Newburgh, New York; E. Fougera & Company, Inc., Hicksville, Long Island, New York; Franklin X-Ray Corporation, Philadelphia, Pennsylvania; General Electric Company, Milwaukee, Wisconsin; Gordon Consultants, New York, New York; Great Books of the Western World, Baltimore, Maryland; Grune & Stratton, Inc., New York, New York; Halsey X-Ray Products, Inc., Brooklyn, New York; Paul B. Hoeber, Inc., New York, New York; Hospital Microfilming Company, Pearl River, New York; Howdon Videx Products Corporation, Mount Vernon, New York; Philip A. Hunt Company, Palisades Park, New Jersey; Ilford, Inc., New York, New York; Keleket X-Ray Corporation, Waltham, Massachusetts; Leishman X-Ray Engineering Corporation, Los Angeles, California; Liebel-Flarsheim Company, Cincinnati, Ohio; Low X-Ray Corporation, New York, New York; Machlett Laboratories, Inc., Springdale, Connecticut; Mallinckrodt Chemical Works, St. Louis, Missouri; Mattern X-Ray Division—Land Air, Incs., Chicago, Illinois; Micro X-Ray Recorder, Inc., Chicago, Illinois; North American Philips Company, Inc., New York, New York; Nuclear-Chicago Corporation, Des Plaines, Illinois; Pako Corporation, Minneapolis, Minnesota; Pennsylvania X-Ray Corporation, Glenside, Pennsylvania; Physicians' Technical Equipment Company, Milwaukee, Wisconsin; Picker X-Ray Corporation, White Plains, New York; Profexray, Inc., Maywood, Illinois; W. B. Saunders Company, Philadelphia, Pennsylvania; Schick X-Ray Company, Inc., Chicago, Illinois; Frank Scholz X-Ray Corporation, Boston, Massachusetts; Siemens New York, Inc., New York, New York; E. R. Squibb & Sons, New York, New York; Standard X-Ray Company, Chicago, Illinois; Charles C Thomas, Publisher, Springfield, Illinois; Thuresson-Angrabright-Bennett, Inc., Skokie, Illinois; United States Radium Corporation, Morristown, New Jersey; Westinghouse Electric Corporation, Pittsburgh, Pennsylvania; Winthrop Laboratories, New York, New York; X-Ray Solutions Services, Detroit, Michigan; Year Book Medical Publishers, Inc., Chicago, Illinois.

NEWS ITEMS

AMERICAN ROENTGEN RAY SOCIETY ELECTS OFFICERS

At the Sixty-second Annual Meeting of the American Roentgen Ray Society held at the Deauville Hotel in Miami Beach, Florida, September 26–29, 1961, the following officers were elected: President, Dr. Traian Leucutia; President-Elect, Dr. Earl E. Barth; First Vice-President, Dr. Ralph M. Caulk; Second Vice-President, Dr. Richard Schatzki; Secretary, Dr. C. Allen Good; Treasurer, Dr. Stephen W. Brown. Dr. Robert C. Pendergrass is the newly elected Chairman of the Executive Council.

The Sixty-third Annual Meeting of the Society will be held October 2–5, 1962, in Washington, D.C.

AMERICAN COLLEGE OF RADIOLOGY CONFERS HONORARY MEMBERSHIP

The American College of Radiology held a special convocation in São Paulo on Wednesday, September 6, 1961 to confer honorary membership on the following: Dr. Luis Arrieta-Sanchez, Editor of *Radiología* of Panamá; Dr. Nicola C. Caminha, President of the Brazil College of Radiology, Rio de Janeiro, Brazil; and Dr. Antonio Pinto-Vieira, Director of the Institute of Cancer, Rio de Janeiro, Brazil.

The convocation was presided over by Dr. Philip J. Hodes. Dr. E. P. Pendergrass acted as Chairman of the Board of Chancellors; Dr. Ross Golden as Sergeant of Arms. Dr. J. A. del Regato read the citations of the new members in Portuguese.

FOURTEENTH ANNUAL MIDWINTER RADIOLOGICAL CONFERENCE

The Fourteenth Annual Midwinter Radiological Conference, sponsored by the Los Angeles Radiological Society, will be held at the Biltmore Hotel, Los Angeles, California, on Saturday and Sunday, February 3 and 4, 1962.

An outstanding program of pertinent interest has been arranged and the guest speakers will be Doctor David Sutton, Saint Mary's Hospital, London, England; Doctor Scott Dunbar, Montreal Children's Hospital, Montreal, Canada; Doctor Richard Marshak, Mt. Sinai Hospital, New York, New York; Doctor Gilbert Fletcher, M. D. Anderson Hospital, Houston, Texas; and Doctor Henry Kaplan, Stanford Medical Center, Stanford, California.

The conference fee of $25.00 includes two luncheon meetings featuring questions and answers. A banquet ($7.50 per plate) preceded by cocktails will be held Saturday evening at the Biltmore Bowl. Reservations may be made through: V. G. Mikity, M.D., 2010 Wilshire Boulevard, Los Angeles 57, California.

Courtesy cards will be available to residents in radiology and Radiologists in the Armed Forces by advance registration, with reduced tariff for luncheons and banquet. Hotel reservations should be made through the Convention Manager, Biltmore Hotel, Los Angeles, California.

UNIVERSITY OF MINNESOTA ANNOUNCES CONTINUATION COURSE

The University of Minnesota announces a Continuation Course in Urologic Radiology for Radiologists, Nov. 6–10, 1961.

Guest participants will include Doctors Guido Currarino, New York; Arthur T. Evans, Cincinnati; John A. Evans, New York; Carl L. Gillies, Iowa City; James O. Hoppe, Rensselaer, New York; John A. Hutch, San Francisco; Harry Z. Mellins, Brooklyn; Olle Olsson, Sweden; Eugene F. Poutasse, Cleveland; Vincent Vermooten, Fort Sam Houston, Texas; and Keith Waterhouse, Brooklyn.

For information write: Director, Center for Continuation Study, University of Minnesota, Minneapolis, Minn.

BOOKS RECEIVED

The Fundamentals of X-ray and Radium Physics. Third edition. By Joseph Selman, M.D., Clinical Assistant Professor of Radiology, University of Texas, Southwestern Medical School; Director, School for X-ray Technicians, Tyler Junior College; Director, Radiology Department, Medical Center Hospital; Attending Staff in Radiology, Mother Frances Hospital; Consultant in Radiology, East Texas Tuberculosis Hospital, Tyler, Texas. Cloth. Price, $8.50. Pp. 364, with illustrations and tables. Charles C Thomas, Publisher, 301–327 East Lawrence Avenue, Springfield, Ill., 1961.

Respiration in Health and Disease. By R. M. Cherniack, M.D., M.Sc., F.R.C.P. (C.), Assistant Professor of Medicine, University of Manitoba School of Medicine; Director, Respiratory Division, Clinical Investigation Unit, and Assistant Physician, Winnipeg General Hospital; Consultant in Respiratory Diseases to Children's Hospital and Municipal Hospitals, Winnipeg, Canada; and L. Cherniack, M.D., B.Sc., M.R.C.P. (Lond.), F.R.C.P. (C.), F.A.C.P., Assistant Professor of Medicine, University of Manitoba School of Medicine; Associate Physician, Winnipeg General Hospital; Physician, Division of Medicine, Winnipeg Clinic, Winnipeg, Canada. Cloth. Pp. 403, with 92 illustrations. W. B. Saunders Company, 218 West Washington Square, Philadelphia, 1961.

Advances in Oto-Rhino-Laryngology. Vol. VIII. Edited by Prof. Dr. L. Rüedi, Zürich, Switzerland. Cloth. Price, $13.00. Pp. 431, with numerous illustrations. S. Karger, Basel, Switzerland. In the U. S. A. and Canada, Albert J. Phiebig, P. O. Box 352, White Plains, N. Y., 1961.

Advances in Tuberculosis Research. Vol. II. Edited by Dr. Hubert Bloch, Pittsburgh, Pennsylvania. Cloth. Price, $18.00. Pp. 282, with illustrations. S. Karger, Basel, Switzerland. In the U. S. A. and Canada, Albert J. Phiebig, P. O. Box 352, White Plains, N. Y., 1961.

Reconstruction Surgery and Traumatology. Edited by E. L. Compere, Chicago, Illinois. Cloth. Price, $11.00. Pp. 139, with many illustrations. S. Karger, Basel, Switzerland. In the U. S. A. and Canada, Albert J. Phiebig, P. O. Box 352, White Plains, N. Y., 1961.

Public Health Papers. No. 6. Ionizing Radiation and Health. By Bo Lindell, Institute of Radiophysics, Stockholm, Sweden; and R. Lowry Dobson, Chief Medical Officer, Radiation and Isotopes, World Health Organization, Geneva, Switzerland. Paper. Price, $1.00 Pp. 81, with many tables. World Health Organization, Geneva, Switzerland. In the U. S. A., Columbia University Press, International Documents Service, 2960 Broadway, New York, N. Y., 1961.

Public Health Papers. No. 9. Teaching of Psychiatry and Mental Health. Contributors: M. Bleuler, O. V. Kerbikov, E. E. Krapf. T. A. Lambo, S. Lebovici, Tsung-yi Lin, I. Matte-Blanco, M. Ostow, T. F. Rodger, J. Stoetzel, and E. S. Turrell. Paper. Price, $2.00. Pp. 186, with tables. World Health Organization, Geneva, Switzerland. In the U. S. A., Columbia University Press, International Documents Service, 2960 Broadway, New York, N. Y., 1961.

Radiographic Anatomy of the Human Skeleton; A Handbook for Radiographers. By W. H. Johnson, F.S.R., Lecturer in Radiographic Anatomy, 1940–1956, for the Society of Radiographers Membership Examination at the Ilford Department of Radiography and Medical Photography, Tavistock House, London; and J. A. Kennedy, M.B., M.R.C.S., D.M.R., D.M.R.D., Lecturer in Radiological Anatomy, King's College, University of London, Senior Radiologist, St. Helier Hospital, Carshalton, England. Cloth. Price, $10.00. Pp. 280, with 233 illustrations. Williams & Wilkins Company, 428 East Preston Street, Baltimore, 1961.

The Year Book of Cancer, 1960–1961. Compiled and edited by Randolph Lee Clark, Jr., B.S., M.D., M.Sc. (Surgery), D.Sc. (Hon.), Director and Surgeon-in-Chief, The University of Texas M. D. Anderson Hospital and Tumor Institute; Professor of Surgery, The University of Texas Postgraduate School of Medicine; Clinical Professor of Surgery, Baylor University College of Medicine; Chairman, Committee on Cancer, American College of Surgeons; F.A.C.S.; and Russell W. Cumley, B.A., M.A., Ph.D., Director of Publications, The University of Texas M. D. Anderson Hospital and Tumor Institute; Professor of Medical Journalism, The University of Texas, Postgraduate School of Medicine; Executive Editor, Medical Arts Publishing Foundation, Houston, Texas. Cloth. Price, $8.50. Pp. 539, with 189 illustrations. Year Book Medical Publishers, Inc., 200 East Illinois Street, Chicago, 1961.

British Columbia Cancer Institute, Vancouver; Five Year Results of Treatment 1946–1954 with a Synopsis of Treatment Policy 1961. By A. Maxwell Evans, M.D., C. M., D.M.R.E., Director, British Columbia Cancer Institute, Clinical Assistant Professor, Department of Surgery, University of British Columbia, Cloth. Pp. 269. British Columbia Cancer Institute, 2656 Heather Street, Vancouver 9, B.C., 1961.

ABSTRACTS OF RADIOLOGICAL LITERATURE

Department Editor: T. LEUCUTIA, M.D., Harper Hospital, Detroit 1, Michigan

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ROENTGEN DIAGNOSIS

Head

Steven, J. L. Some pitfalls in the angiographic diagnosis of intracranial tumour pathology. *Clin. Radiol.*, July, 1961, *12*, 194–198. (From: West of Scotland Neurosurgical Unit, Killearn Hospital, Glasgow, Scotland.)

Angiographic diagnosis of the nature of a tumor is now being readily accepted by neurologists and neurosurgeons, throwing a correspondingly greater burden on the radiologist. The author gives 3 case histories in which the angiographic appearances were unusual.

Case 1. A diagnosis of meningioma was made on the basis of the angiographic findings. Complete occlusion of the left internal carotid artery at its origin was demonstrated, and there were two areas of abnormal vasculature in the superficial left frontoparietal region. At craniotomy no tumor was found. The misleading appearance may have been due to some form of anastomotic circulation via the superficial temporal artery and the middle meningeal artery, but this is mere speculation.

Case 2. Right common carotid angiography showed mid-line vessels displaced to the left, and several small wooly opacities of different sizes appeared in the late arterial and capillary phases. These had cleared in the venous phase. A diagnosis of multiple tumors was suggested. At postmortem examination there were multiple hemorrhagic infarcts corresponding in position to the opacities seen on the roentgenograms. Presumably the opacities were due to contrast medium circulating through numerous dilated capillaries adjacent to the ischemic zones.

Case 3. A left carotid angiogram revealed a small knot of vessels in the posterior temporal region with an early filling vein draining the area. This was interpreted as a small arteriovenous malformation. Five weeks later a repeat left angiogram revealed a marked shift of mid-line vessels to the right, elevation of the middle cerebral group of vessels, and a large area of poorly defined tumor vessels. The patient expired seven days later. A postmortem examination was not made, but the roentgenographic appearance indicated a rapidly growing glioblastoma.—*Samuel G. Henderson, M.D.*

Tori, G., and Garusi, G. F. Left carotid-jugular arteriovenous fistula; angiocardiographic study before and after surgical operation. *Radiol. clin.*, 1961, *30*, 76–85. (Address: G. Tori, Institute of Radiology and Radium Institute of the University, Bologna, Italy.)

This is a case report of a five year old boy with a congenital left carotid-jugular arteriovenous fistula. This had caused a large left to right extracardiac shunt and had resulted in cardiomegaly and heart failure.

On angiocardiography the left common carotid was dilated to three times normal size and appeared to be twisted. There were two large shunts (0.5 cm.) between the left common carotid and the external jugular vein, one near the angle of the jaw and the other 4 cm. lower. The left innominate vein was markedly dilated and tortuous. At surgery, the abnormal shunts were removed and there was immediate improvement in the patient's symptoms.

Three years after surgery a chest film disclosed marked reduction in the size of the heart. An angiocardiogram showed that the left common carotid was still considerably enlarged but no shunt was present.—*J. L. Williams, M.D.*

Tindall, George T., and Cupp, Horace B., Jr. Vertebral arteriography by retrograde injection of the right common carotid artery. *Radiology*, May, 1961, *67*, 742–747. (Address: G. T. Tindall, Department of Surgery, Duke University Medical Center, Durham, N. C.)

The authors describe a simple, percutaneous method for obtaining vertebral arteriograms during the course of carotid arteriography. The basic feature of this method is that a thin-wall 17 gauge needle is inserted into the right common carotid artery, below the carotid bifurcation, with the point directed toward the heart. An injection of 20 cc. of 50 per cent hypaque is made manually in a retrograde direction with compression on the common carotid artery above the needle and on the ipsilateral brachial artery. Exposures are made at the completion of the injection, producing diagnostic vertebral arteriograms in the majority of cases. A slower injection of 10 cc. of 50 per cent hypaque, without vessel compression, allows direct right carotid arteriography.

Critical analysis of 125 injections in a group of 60 patients revealed diagnostic vertebral angiograms in 42 patients, or 70 per cent. The majority of failures was due to improper timing or incomplete compression of the distal carotid artery. This latter observation prompts the recommendation that the same concentration of contrast medium used for carotid arteriograms should be used for the retrograde vertebral arteriograms. Retrograde injections were made into the left common carotid artery in 6 patients without visualization of either the left vertebral or subclavian arteries.

The two chief advantages of the retrograde method are that it avoids direct puncture of the vertebral artery and can be accomplished under local anesthesia. It is especially worthy of consideration in patients with thrombosis of the internal carotid artery since it avoids the risk of threading

the tip of the needle into the involved portion of the vessel. Although the one "blind spot" in this technique is the failure to visualize the left vertebral artery, a complete survey of the intracranial arterial circulation can be accomplished in one session, through bilateral carotid artery punctures.—*Edward B. Best, M.D.*

OSTROWSKI, A. Z., HARDY, W. G., LINDNER, D. W., THOMAS, L. M., and GURDJIAN, E. S. Retrograde brachial vertebral-basilar angiography; an analysis of angiographic visualization of the vertebral-basilar system and branches. *A.M.A. Arch. Neurol.*, 1961, *4*, 608–616. (Address: E. S. Gurdjian, Wayne State University, 1401 Rivard St., Detroit 7, Mich.)

Extracranial vascular occlusive lesions in the stroke syndrome were investigated in 90 patients by the effective and safe method of retrograde brachial angiography. In the earlier cases, open arteriotomy and general anesthesia was practiced; later, percutaneous technique with a 17 gauge spinal needle in each brachial artery was successful. About 20 cc. of 50 per cent hypaque was used for each of the 4 to 6 manual injections required to obtain angiograms in varying projections. (It is of note that "as much as 300 ml." of the opaque medium was employed in some cases!) No important complications occurred.

The technique produced visualization of the right carotid in 75 cases; of the left carotid in 3 cases. Opacification of the left carotid occurred only with injection of the right brachial and success seemed dependent on an anomalous origin from a common innominate. Both vertebrals (when patent) were opacified in all cases. In only a third of the carotids shown could the origin of the vessel be seen; the origin of the vertebrals was seen in the majority. In over half of the group, the origin of the posterior inferior cerebellar artery was shown. Evaluation of the basilar arteries was often permitted by this method.

Artifact or anomaly may be suspected when a portion of the vertebral-basilar system is not opacified, but in the majority this sign indicates disease. Left and right vertebrals are rarely of the same caliber and nonfilling of the intracranial portion of the vertebral artery was usually interpreted as artifact if it was on the side of the smaller vertebral and if the basilar artery filled from the opposite larger vertebral. Among those suspected of vascular occlusive disease the procedure strongly supported this diagnosis (*i.e.*, 77 per cent showed atheromatous abnormalities.)

Finer points of technique are not described but references are supplied and should be consulted. When occlusive disease of the vertebral-basilar system is suspected this would seem to be the diagnostic procedure of choice because of its safety and reliable graphic result. The hemorrhage and pneumothorax which may attend the percutaneous subclavian approach are avoided. The important site, the origin of the vessel, is usually demonstrated as it often is not in a percutaneous vertebral artery puncture. One should not anticipate opacification of the left carotid by this method and poor visualization of the origin of the right carotid is a deficiency shared by most techniques. The role played by hydrodynamics to prevent opacification of portions of the system and thus mimic structural occlusion is the *bête noire* of angiography of the posterior fossa and will help to keep medicine an art.—*Jack Reynolds, M.D.*

OLLER, JOSE L. GARCIA, NELSON, N., ESPENAN, P., FERNANDEZ, W., MATHIRNE, C., and HAYDEL, W. Angiography by brachiovertebral and temporal arterial catheter. *South. M. J.*, Apr., 1961, *54*, 345–349. (From: The Department of Neurosurgery, Mercy Hospital, and the Independent Neurosurgery Service, Charity Hospital, New Orleans, La.)

The authors summarize their results in angiography of the cerebral vessels by direct arterial catheterization. Brachial angiography makes possible routine studies of the vertebral circulation with freedom from spasm and direct injury plus visualization of the carotid artery. The authors also utilized catheterization of the temporal arteries and occasionally they combined the procedures by using the right brachial and left temporal arteries in one examination. This combined procedure produced an excellent one stage cerebral angiogram.

They conclude that catheterization of the brachial artery is safe and reliable for routine vertebral angiography, as is temporal catheterization for carotid angiography, with the latter also being an excellent technique for pediatric patients. They found that critically ill patients with head injuries and subarachnoid hemorrhage are accurately and safely studied with catheterization techniques. Also, vertebral angiography gave essential diagnostic information in one-third of the cases of brain tumors studied.—*Richard L. Jontz, M.D.*

LAMB, JOHN, and MORRIS, LEON. The carotid-basilar artery; a report and discussion of five cases. *Clin. Radiol.*, July, 1961, *12*, 179–186. (From: X-ray Department, Brook Hospital, Shooters Hill Road, London, S.E. 18, England.)

An anomalous vessel joining the carotid and basilar arteries has been known to anatomists since it was first described by Quain in 1844. The first case diagnosed during life by angiography was described by Sutton in 1950.

The authors give 5 case histories in which this

anomalous vessel was found by arteriographic examination. They state that their experience and a study of the literature show that, in almost all cases, the anomaly is found incidentally during the course of routine angiography. Of their 5 patients, the first had a metastatic cerebral tumor, the second was angiographically normal apart from the anomalous vessel, the third had an intracerebral hematoma associated with a hemisphere angioma, the fourth presented with subarachnoid hemorrhage from an aneurysm on the posterior communicating artery of the opposite side, and the fifth case had an extensive infiltrating glioma.

These 5 cases were encountered in the course of approximately 1,500 arteriographies. Other investigators have found this anomaly with slightly less or slightly greater frequency; hence, it is not excessively uncommon.

Roentgenologically, a full-axial view is recommended in trying to demonstrate the exact appearance of the anomalous vessel. With a half-axial view the apex of the basilar artery and the vessels arising therefrom are obscured by the anterior cerebral artery.—*Samuel G. Henderson, M.D.*

Begg, A. C. Radiographic demonstration of the "hypoglossal artery"; a rare type of persistent anomalous carotid-basilar anastomosis. *Clin. Radiol.*, July, 1961, *12*, 187–189. (From: The Department of Diagnostic Radiology, Dunedin Hospital, and The Otago Medical School, Dunedin, New Zealand.)

The basilar artery may arise from the posterior aspect of the cervical portion of the internal carotid artery. This represents a persistence of certain small anastomotic channels linking the internal carotid and vertebral arterial systems in the embryo. Most writers have considered that this vessel represents the hypoglossal artery. It has been described previously on four occasions as a finding in anatomic dissections.

The author, discovered this anomaly on arteriography in a patient who later came to surgery for an inoperable brain tumor. This is apparently the first time that this anomalous artery has been found during life.—*Samuel G. Henderson, M.D.*

Salinas, F. de los Santos, and Gayarre, M. Gil. La sialografía. Tecnica, indicaciones y resultados. (Sialography. Technique, indications and results.) *Acta ibér. radiol. cancerol.*, Apr.-June, 1960, *15*, 107–116. (From: Catedra de Radiología y Terapeutica Fisica de la Universidad de Madrid, Spain.)

The technique for contrast roentgenography of the parotid and submaxillary glands is described with great detail. The patient is asked to rinse his mouth with warm water (40°C.) in order to relax and dilate the distal end of the excretory ducts. The duct is dilated, if necessary, and catheterized with a blunt-end needle not more than 1 mm. in diameter. For a contrast medium, the authors much prefer iodized oils to aqueous media. They inject 1.5 to 2 cc. in the submaxillary gland and 2.5 to 3 cc. in the parotid until the patient experiences pain and distention of the gland is noted. With the needle in place to prevent reflux, roentgenograms are taken. The needle is withdrawn and excretion of the contrast medium is achieved by having the patient chew gum and rinse the mouth to eliminate the contrast medium in the oral cavity. Additional roentgenograms are obtained during this time. The best projection for visualization of the submaxillary gland is obtained with the roentgen-ray beam angled 60° from the vertical plane and the patient lying supine with the head rotated to the side being studied. For study of the parotid gland, lateral views are preferred.

The authors have found interesting features in cases of transparent calculi, chronic inflammations, fistulae, relapsing parotitis, Mikulicz's disease, benign and malignant tumors, actinomycosis, and in patients who have received irradiation for conditions in the region of the salivary glands.—*F. Comas, M.D.*

Neck and Chest

Rogan, J. M., Ashford, J. R., Chapman, P. J., Duffield, D. P., Fay, J. W. J., and Rae, S. Pneumoconiosis and respiratory symptoms in miners at eight collieries. *Brit. M. J.*, 1961, *1*, 1337–1342. (From: Pneumoconiosis Field Research, National Coal Board, London, England.)

This report on a project which was started in 1953 includes an investigation into the correlation of coal dust, pneumoconiosis, respiratory diseases, forced expiratory volume ($F.E.V._1$), and roentgen findings.

A total of 9,758 men from eight British collieries were examined. The prevalence of pneumoconiosis increased with age. It varied from 9 to 41 per cent from one colliery to another.

Prevalence of respiratory symptoms increased with age also. The mean $F.E.V._1$—that is, the volume of air expired during the first second of a forced expiration after a full inspiration—in men with respiratory symptoms was about .2 to .4 liter less than that in men without respiratory symptoms.

There was some relation between pneumoconiosis and respiratory symptoms. Respiratory symptoms were associated with a smaller fall in the $F.E.V._1$ in men with pneumoconiosis than in men with normal roentgenograms.

Irrespective of the presence or absence of pneumoconiosis and respiratory symptoms, the $F.E.V._1$ fell uniformly with age.—*David C. Alftine, M.D.*

Robertson, A. John, Rivers, D., Nagelschmidt, G., and Duncumb, P. Stannosis; benign pneumoconiosis due to tin dioxide. *Lancet*, 1961, *1*, 1089–1093. (Address: A. J. Robertson, Royal Infirmary, Liverpool, England.)

In 1955, 121 patients were studied roentgenographically and were found to have peculiar densities on chest roentgenograms. The diagnosis was not confirmed, but was thought certainly to be a benign pneumoconiosis due to tin dioxide.

The lungs of 7 of these patients have been obtained from necropsy. This is a report of the findings after pathologic examination, chemical and x-ray-diffraction analysis, and x-ray-emission spectroscopy.

Although tin in various compounds is processed by these workers, it appears that when the ore is reduced and smelted, the hot gases from the molten metal are inhaled. This is thought to be the main cause of "stannosis" in the lungs.

In the 7 patients observed, there was no significant pulmonary fibrosis and no evidence of silicosis. Chemical and x-ray-diffraction studies showed that the lungs contained tin dioxide.

The authors confirmed the diagnoses, made six years previously, of benign pneumoconiosis due to tin dioxide.—*David C. Alftine, M.D.*

Semple T., and Lancaster, W. M. Noisy pneumothorax; observations based on 24 cases. *Brit. M. J.*, 1961, *1*, 1342–1346. (From: Victoria Infirmary, Glasgow, Scotland.)

A total of 24 patients with noisy pneumothorax in the past 10 years has been encountered. The patients have presented chest findings of clicking, crackling, or crunching sounds at the cardiac apex with left-sided pain.

The youngest were 4 youths of eighteen years. The oldest was a woman of thirty-six. Pain was severe initially, but gradually lessened within hours, while the noise often lasted days.

Eighteen patients had obvious pneumothorax on roentgenograms. In 4 others special studies were required in order to demonstrate the condition roentgenographically. In 2 patients no abnormality was noted, but expiratory roentgenograms were not taken.

Hamman's sign may be heard in traumatic or severe spontaneous mediastinal emphysema, but shallow left pneumothorax is much more frequently the cause. The authors list several serious conditions requiring differential diagnosis.

Noisy pneumothorax is a benign condition, usually occurring in young healthy males, and is invariably left-sided. In a healthy patient with sudden chest pain this diagnosis cannot be excluded unless a roentgenogram is taken in full expiration.—*David C. Alftine, M.D.*

Smart, M. J., and Spencer, E. W. Bronchiolar ectasia and bronchial gland dilatation. *J. Canad. A. Radiologists*, 1961, *12*, 56–62. (From: University Hospital, Saskatoon, Sask., Canada.)

The authors take exception to the idea "suggested in recent American literature" that bronchiolar ectasia and bronchial gland dilatation are uncommon. In a review of 161 consecutive bronchograms, 36 cases of mucous gland dilatation and 10 cases of bronchiolar ectasia were found. In 2 of the cases demonstrating both abnormalities, bronchiectasis was also present.

"Trapping" of contrast material (aqueous dionosil) within ectatic cavities on days subsequent to bronchography was found to be of diagnostic value in doubtful cases of bronchiolar ectasia.

The authors feel that the presence of both bronchiolar ectasia and mucous gland dilatation in the absence of bronchiectasis justifies the diagnosis of chronic bronchitis.—*W. C. MacCarty, Jr., M.D.*

Cornudella, R., Frechilla, L., Lorenzo, T., Margarit, F., Conti, J. Reventós, and Albertí, A. La función ventilatoria en las bronconeumopatías congénitas; estudio de 5 casos. (Pulmonary ventilation studies in broncho-pulmonary congenital anomalies; study of 5 cases.) *Ann. med.*, Oct., 1960, *46*, 516–530. (From: Departamento de Exploración Funcional del Servicio de Aparato Respiratorio del Hospital de la Santa Cruz y San Pablo de Barcelona, Barcelona, Spain.)

Five cases are reported with complete clinical, roentgenologic and pulmonary function studies. The cases include 2 aplasias of the left lung, a congenital cyst of the left lung secondarily infected with tuberculosis, a polycystic left lung and a congenital bronchiectasis of the left lower lobe.

It was found that there was a deficit in the pulmonary function in all cases. The deficit appears to be directly proportional to the precocity of the malformation and the amount of pulmonary tissue involved. The uninvolved lung appears to compensate partially by using its pulmonary reserve and increasing its output. This compensation appears to be about 15 to 30 per cent.—*Cesar E. Rosa Perez, M.D.*

Dragoni, G., and Galli, G. Le manovre di Valsalva e di Müller e la loro utilità nello studio radiologico di alcune manifestazioni patologiche del torace. (The utility of the Valsalva and' the Müller maneuvers in the radiological study of thoracic pathologic

manifestations.) *Radiol. med.*, Mar., 1961, *47*, 193–207. (Address: G. Dragoni, Orio-Litta, Milano, Italy.)

The Valsalva maneuver consists of a prolonged forced and blocked expiration. The Müller maneuver is the inverse of the preceding and consists of a forced and blocked inspiration.

During the Valsalva maneuver there is: (1) an increase in pulmonary transparence; (2) a better definition of the arterial tree; (3) reduction of the venous perihilar shadows; (4) a reduction or loss of fogging because of the diminished filling of the alveolar capillaries.

The Müller maneuver is not particularly useful in the roentgenologic diagnosis of pulmonary parenchymal pathologic manifestations. It may, however, have value in the differentiation of pulmonary vascular from parenchymal lesions.

The Valsalva maneuver was adopted for: (1) the demonstration of parenchymal metastasis which might be obscured by the vascular pattern; and (2) the recognition of hilar and mediastinal lymphadenopathy which is rendered difficult, by the superimposition of vascular shadows.—*A. A. Blasi, M.D.*

Hemley, Samuel D., Arida, Edward J., Diggs, A. Monroe, and Freeman, Sumner L. Percutaneous cricothyroid membrane bronchography. *Radiology*, May, 1961, *76*, 763–769. (Address: S. D. Hemley, St. Luke's Hospital, Amsterdam Ave. and 113th St., New York 25, N. Y.)

This study is based on six months' experience with 42 consecutive transcricoid needle punctures for bronchographic examinations. The technique is described in detail. There were only three complications directly related to the procedure. In one patient subcutaneous emphysema developed but cleared in three days. A second patient experienced hoarseness for one day. A third patient developed a brisk hemoptysis presumably from severe tuberculous bronchiectasis, and death from cardiac arrest ensued during administration of anesthesia for bronchoscopy on the same day.

With absorbable contrast medium, antibiotic control, and careful attention to asepsis, the method appears to be safe, effective, and time saving, particularly for departments of radiology in large and busy general hospitals. The frequency and seriousness of the complications of this procedure appear to have been exaggerated in the past.

Since submission for publication, 43 additional studies have been performed with one minor complication, subcutaneous emphysema of the neck discovered on a routine 24 hour post-drainage study.—*Walter H. Jarvis, Jr., M.D.*

Garusi, G. F. Opacification of the bronchial arteries in the living; (a new method of experimental demonstration). *Radiol. clin.*, 1961, *30*, 65–75. (Address: Istituto del Radio dell'Università, Policlinica San Orsola, Bologna, Italy.)

Opacification of the bronchial arteries during angiocardiography or aortography is rare and rather poor. Visualization of the bronchial arteries appears to be limited to cases in which they have become dilated in response to the diminished pulmonary arterial circulation which occurs in chronic lung disease or cardiovascular disease.

The author has achieved better visualization of the bronchial arteries in dogs by performing retrograde thoracic aortography during acetylcholine induced cardiac arrest. He concludes that further experiments are necessary to define the possibilities and the practical importance of the method.—*J. L. Williams, M.D.*

Ormond, Robert S., Poznanski, Andrew K., and Templeton, Arch W. Pulmonary veins in congenital heart disease in the adult. *Radiology*, June, 1961, *76*, 885–893. (Address: R. S. Ormond, The Henry Ford Hospital, Detroit 2, Mich.)

The routine roentgenogram of the chest remains the base line for appraisal of the cardiac patient. The usual lesion encountered in congenital heart disease in the adult is monopathic; therefore, the radiologist can be quite accurate in his prediction of the abnormality. The present study is directed to an analysis of the pulmonary veins in congenital heart disease in adults, since the pulmonary veins reflect the flow of blood through the lung fields.

Four major venous channels are constant in position and can usually be identified on routine posteroanterior roentgenograms of the chest. They are: posterior division of the superior pulmonary veins, the superior and inferior divisions of the inferior pulmonary veins and the middle pulmonary veins. These vessels can be seen crossing the arterial channels and entering the left atrium below the main pulmonary arteries. No value can be ascribed to the diameter of a normal vein, because there is considerable variation in vein size in normal patients. With experience a reasonably accurate appraisal of the deviations from normal is possible. The authors utilize a classification which separates the veins into five categories: (1) definitely reduced; (2) reduced to normal; (3) normal; (4) normal to increased; and (5) definitely increased. This is a gross division and subject to inexactness but it is of value when the appearance of the veins is correlated with the arterial and the cardiac configuration.

Eighty-six patients were included in this study. Right heart catheterization was done in all, with determination of intracardiac, pulmonary artery, and pulmonary wedge pressures. The oxygen saturation

of the blood was determined at suitable levels in the heart and pulmonary artery. Ten of the patients had undergone selective angiocardiography and in 43 the lesions were confirmed at the time of corrective surgery or at autopsy.

In lesions such as pulmonary stenosis and pulmonary hypertension, leading to shunt balance and reversal, in which the cardiac output is reduced, the veins are small. In uncomplicated interatrial septal defect, the pulmonary veins are definitely increased in size. In patent ductus arteriosus with left-to-right shunt the veins are large. In cases with a balanced or a reversed shunt, the veins are smaller than those seen in normal patients and may approach the size of veins in severe pulmonary stenosis. In patients with pulmonary hypertension, a successful surgical result depends upon the presence of a significant left-to-right shunt. Vein size offers one more method of evaluating the amount of such a shunt.—*A. W. Sommer, M.D.*

Levin, Bertram, and White, Harvey. Total anomalous pulmonary venous drainage into the portal system. *Radiology*, June, 1961, *76*, 894–901. (Address: B. Levin, Michael Reese Hospital, Chicago 16, Ill.)

Although many variations of anomalous pulmonary venous drainage are known, the majority is supradiaphragmatic with the veins emptying into the right side of the heart directly or via interposed vascular channels. Some of these can be suspected from the characteristic roentgenographic contour of the heart and great vessels. Total anomalous pulmonary venous drainage into infradiaphragmatic vessels is far less common, comprising 10 to 20 per cent of all cases, and the drainage is usually into the portal vein or ductus venosus. These patients, however, present a characteristic and distinctive roentgenographic appearance of the pulmonary vascular pattern, rather than the heart or great vessels, which permits accurate diagnosis.

Four cases of infradiaphragmatic total anomalous pulmonary venous return are presented, each with similar characteristic pulmonary vascular markings. All were cyanotic white male infants who expired at approximately two months of age. Diagnosis was established by angiocardiography in one case, and the 3 additional cases were proved by autopsy. In each instance the pulmonary veins joined to form a single trunk, which traversed the diaphragm either through the esophageal hiatus or a special hiatus just anterior to the esophagus, and emptied into either the portal system (3 cases) or the ductus venosus (1 case). The expected shunt was provided by a large ventricular septal defect in 1 case and a patent ductus arteriosus in the other 3. Two of the cases with a patent ductus also presented a patent foramen ovale.

The interesting and characteristic feature in each instance is a prominent mottled and finely reticulated appearance of the pulmonary vascular markings which extends to the periphery of the lung fields. Kerley lines can be identified in the lung bases, with varying degrees of clarity. These lines often are more apparent in the lateral view than in the frontal. In all cases the cardiac size and contour are not appreciably altered from the normal which differs from supradiaphragmatic anomalous drainage and other congenital cardiovascular diseases with pulmonary vascular engorgement. In 2 cases the antemortem diagnosis was made from the chest roentgenograms.

The authors, in conclusion, state that this anomaly might be amenable to surgical correction. Although the pulmonary venous trunk is a thin walled channel with varying degrees of scarring, it could be transplanted into the small left atrium which is known to be atrophic and not aplastic.—*Edward B. Best, M.D.*

Anderson, I. M., and Coles, H. M. T. Endocardial cushion defects. *Brit. M. J.*, Mar. 11, 1961, *1*, 696–705. (From: The Westminster Hospital, London, England.)

Misdiagnosing an endocardial cushion defect as a simple ostium secundum patency may be serious because the former cannot always be corrected surgically without the use of extracorporeal circulation. The authors quote Bailey *et al.* and Watkins and Gross as stating that 10–15 per cent of cases operated for atrial septal defects have this more complicated lesion.

They briefly discuss the embryology and subscribe to the terminology used by Campbell and Missen. Thus, a Grade I defect includes persistence of the ostium primum with splitting of the anterior mitral cusp or the septal cusp of the tricuspid value, or both; Grade II includes, with the ostium primum, bifid atrioventricular valve cusps and a high ventricular septal defect; and Grade III includes those with persistence of the common atrioventricular canal.

Symptoms include failure to thrive, dyspnea, respiratory infections and, commonly, cyanosis and heart failure. These occur fairly early and the prognosis is poor. A widely radiating holosystolic murmur is the rule with a mid-diastolic murmur often associated; a thrill may be present.

The authors feel that chest roentgenography often fails to differentiate these lesions from other septal defects with a predominant left to right shunt. They consider that the electrocardiographic pattern is highly characteristic and that catheterization is helpful. Angiocardiography is a definite diagnostic aid. They present 10 cases of this lesion with 2 cases of large ostium secundum defects which were misdiagnosed as the above. Of the 10 cases, 5 died (without surgery) of pneumonia and/or congestive failure. Of 5 which were surgically corrected, 3 are

living and well. Both ostium secundum defects were repaired with 1 of the patients dying.—*William H. Baker, M.D.*

Genitourinary System

Don, Conway. Asymmetry of the kidneys in disease of the main renal artery. *J. Canad. A. Radiologists*, Mar., 1961, *12*, 15–21. (From: The Ottawa General Hospital, Ottawa, Ontario, Canada.)

In a series of 100 normal intravenous pyelograms in patients with normal blood pressures, the author attempted to determine what degree and type of asymmetry can be accepted as a normal variant. In contrast to the normal cases, there were 6 cases with asymmetry in which disease of the main renal artery was shown by aortography at operation or at postmortem examination.

It is recognized that there is normal asymmetry of the kidneys, the right kidney being shorter and thicker than the left. In addition to evaluating the length of the kidneys, the author determined the thickness of the parenchyma by measuring from the tips of the papillae to the nearest portion of the renal outline, at midlevel.

It was found that in 73 per cent of the cases there was less than 1 cm. difference in length between the kidneys of the same subject. In 27 per cent there was asymmetry of 1 cm. or over. It was decided that asymmetry in length alone was of no significance. In the control series, when the kidney was 1 cm. or more shorter, the renal parenchyma was usually thicker. It was therefore concluded that, if asymmetry in length is accompanied by parenchymatous thinning of the shorter kidney of 3 mm. or more, this is suggestive evidence of main renal artery disease, and an indication for aortography. It is important, however, to be aware of the fact that absence of these findings does not preclude a renal artery lesion.—*J. N. Ané, M.D.*

Sutton, David, Brunton, F. J., and Starer, F. Renal artery stenosis. *Clin. Radiol*, Apr., 1961, *12*, 80–90. (From: Diagnostic X-ray Department, St. Mary's Hospital, London W.2, England.)

Renal artery stenosis is now recognized as a significant cause of hypertension. In over 1,000 examinations by aortography for conditions such as aortic thrombosis, iliac stenosis or abdominal aneurysm, the authors have seen 20 cases of renal artery stenosis, usually as a chance finding. In this group, 11 individuals were known to have hypertension and 5 were known to be normotensive. As part of the investigation of 260 known hypertensives who were subjected to arteriography for the purpose of finding or excluding renal artery stenosis or other vascular disease, 26 cases of renal artery stenosis were encountered. Nine cases of stenosis were shown by selective renal arteriography and 17 by direct aortography.

Aortography may be performed using three different techniques. The first is simple translumbar aortography; the second is aortography by percutaneous transfemoral catheterization; and the third is percutaneous selective renal aortography in which the opaque catheter tip is guided into the renal artery under fluoroscopic control. The particular method used is largely a matter of personal experience and preference. The selective method undoubtedly produces the most detailed demonstration of the renal vascular tree with minimal injections of contrast material, but it may be technically impossible to perform in cases of multiple renal arteries.

The most serious side effects reported in the literature have been renal damage and paraplegia, and there is little doubt that most of them have been due to excessive doses of contrast medium. The authors use 20 cc. of 76 per cent urografin as a maximum dose for free injections into the aortic lumen, and it is possible to obtain adequate visualization with lower doses. For selective renal angiography they generally inject 5 cc., or less, of 45 per cent hypaque or 60 per cent urografin. Great caution is necessary in the presence of impaired renal function, either bilateral or unilateral. The hypertension itself is an added hazard as it increases the tendency to hemorrhage and to the formation of large hematomas.

The highest incidence of renal artery stenosis is in patients over forty years of age. The increased frequency in males reflects the predominance of atheroma in men.

The lesions have been grouped into four grades of severity, determined by the degree of encroachment on the arterial lumen as seen on the angiograms. The stenosis is usually shown as a smooth narrowing of the lumen of the artery, and is nearly always less than 1 cm. in length, usually only 2 to 4 mm. The presence of post-stenotic dilatation of the renal arteries was noted 19 times. The stenosis occurred at the origin of the renal artery or within 1 cm. of it in 36 instances. Good demonstration of the origin of the renal arteries will show or exclude the majority of stenoses, although the origin of one or other of the renal arteries is frequently obscured by the superior mesenteric artery.

Certain clinical features may suggest the presence of renal artery stenosis: (a) unexplained hypertension in a young adult; (b) the sudden onset of malignant change in a patient known for years to be a hypertensive; (c) hypertension occurring in a patient with known general atheromatous disease; and (d) the presence of an upper abdominal bruit in a hypertensive patient.

The importance of changes on the plain roentgenogram and pyelogram in cases of renal artery stenosis is stressed. The affected kidney is smaller than the contralateral one. The contrast material may be in greater concentration on the affected side, and its

excretion may be demonstrably less in quantity. The calyces may appear relatively "spastic" on the affected side. This last-mentioned finding is frequently seen on intravenous pyelograms. When it occurs bilaterally, the most common cause is considered to be overenthusiastic dehydration of the patient in the preparation for pyelography. It reflects the low urinary output, and the appearance is due to underfilling of the calyces rather than to "spasm." When seen on one side only, it may be due to other causes, such as inflammatory lesions and calculi.

In 1 case seen on referral, renal artery stenosis on the left side was suggested because of poor contrast on this side together with an alleged left-sided bruit. Arteriography showed a normal left renal artery and stenosis of the right renal artery. Repeat intravenous urographic examination after strict water deprivation for twelve hours showed a much less marked difference in the contrast values between the two kidneys. The appearances described are clearly due to the fact that the ischemic kidney continues to excrete a concentrated urine under conditions of water load, while the contralateral kidney produces a very dilute urine.—*Samuel G. Henderson, M.D.*

Palubinskas, A. J., and Wylie, Edwin J. Roentgen diagnosis of fibromuscular hyperplasia of the renal arteries. *Radiology*, Apr., 1961, *76*, 634–639. (Address: A. J. Palubinskas, Department of Radiology, University of California Medical Center, San Francisco 22, Calif.)

Fibromuscular hyperplasia of the renal artery has recently been reported as a cause of renal artery stenosis and secondary hypertension. The etiology of the lesion is unknown. It has been suggested that the hyperplasia may be secondary to chronic stretching of the renal artery in patients with kidneys that are more mobile than usual.

Renal arteriography is the definitive diagnostic examination. The involved artery presents a distinctive appearance at arteriography. It may have an irregular appearance resembling a string of beads; constricted areas may be single but are more often multiple; post-stenotic dilatation of the renal artery or its branches may occur, with the occasional formation of a berry aneurysm. Fibromuscular hyperplasia appears to involve primarily the middle and/or distal third of the renal artery and may extend into the proximal branches of the main renal artery.

The authors report 11 patients with fibromuscular hyperplasia of the renal arteries seen within a two year period. The patients were all females between fifteen and forty-nine years of age, with hypertension known to have been present one week to twelve years. To date, 9 of the patients have been explored surgically. In each, the stenotic lesion in the renal artery was found to be due to hypertrophy of the muscular elements and, to a lesser extent, to the fibrous connective tissue elements of the media of the vessels. Arterial reconstructive procedures were performed in all cases except 1 in which nephrectomy was done. Following operation, the blood pressure returned to normal levels in 5 and decreased significantly in 3 patients.—*Arno W. Sommer, M.D.*

Witcofski, Richard L., Whitley, Joseph E., Meschan, I., and Painter, William E. A method and parameters for the analysis of renal function by external scintillation detector technic. *Radiology*, Apr., 1961, *76*, 621–627. (Address: R. L. Witcofski, Bowman Gray School of Medicine, Winston-Salem 7, N. C.)

With the advent of ortho-iodohippuric acid I^{131} (OIHA), better scintillation renograms have been obtained. It is now possible to compare right and left curves with greater confidence, and theoretically to quantitate renal function. Various methods of analysis are possible.

The authors' method included renography, blood and bladder activity studies at thirty minutes, and a chart recording of blood activity. The main and plateau times of the vascular phase are determined, as are half-secretory and excretory times; and blood clearance can be calculated.

Details of equipment and techniques are presented.

Parameters expressing the rates of secretion, excretion, and total renal function are given, as well as the normal values, including those for clearance studies.—*James C. Moore, M.D.*

Damanski, M. Cysto-urethrography in paraplegia: its practical application. *Brit. J. Urol.*, Mar., 1961, *33*, 67–75. (From: The Liverpool Regional Paraplegic Centre, Promenade Hospital, Southport, England.)

Paraplegics are very prone to urinary difficulty and cystourethrography is a valuable means of investigating the cause of urinary retention.

The author describes his technique. The bladder is emptied with a catheter and 8 fluid oz. of 10 per cent sodium iodide solution are injected, using a Riches syringe. One anteroposterior and 1 oblique roentgenogram are taken. Following this, a few ounces of the same contrast material are injected into the external meatus and anteroposterior and oblique roentgenograms are obtained; this study visualizes the urethra. Finally, a roentgenogram is taken of the upper urinary tract to investigate reflux.

The author believes that any significant obstruction of the bladder neck or urethra will be demonstrated by this examination. Different sites of obstruction between the bladder neck, external sphincter, and posterior urethra are visualized. If the patients do not improve following surgery, the examination is repeated.

Other abnormalities may be seen, such as diverticula of the urethra or bladder and other evidences of obstruction and infection.

The author feels that this is a very valuable examination in the urologic work-up of paraplegics.—*George L. Sackett, Jr., M.D.*

Kirkland, J. A., and Haslock, Margaret Rae. Transient proteinuria; following intravascular injection of contrast media. *Lancet*, Apr. 1, 1961, *1*, 693–695. (From: The Department of Pathology, University of St. Andrews [Queen's College], and the Royal Infirmary, Dundee, England.)

Because of numerous reports of renal complications, including massive albuminuria following aortography, the authors investigated the occurrence of proteinuria in 53 patients subjected to intravenous pyelography and 14 to aortography.

Results after Intravenous Pyelography. Proteinuria before injection was found in 16 patients and, of these, 3 became more positive following injection; the remainder were unchanged. Seven of the remaining 37 developed proteinuria, most of them to a slight degree varying from 30 mg./100 ml. to 300 mg./100 ml. One had massive proteinuria of 1,000 mg./100 ml. This proteinuria lasted for two days in most patients, but in 2 it persisted for four days.

Results after Aortography. The first 4 patients were examined by translumbar injection with 70 per cent pyelosil and all of these showed a massive proteinuria—from 730 mg. to 1,300 mg./100 ml. The remaining 10 cases were examined by retrograde catheterization of the femoral artery using 70 per cent urokon as a contrast medium and only 1 of these patients had proteinuria, which was slight (100 mg./100 ml.).

The method of testing for protein in the presence of radiopaque contrast material is discussed and the authors recommend the paper strip tests "Albustix" and "Uristix," which do not give false positive results under these conditions.—*George L. Sackett, Jr., M.D.*

Skeletal System

Dahlin, David C., Coventry, Mark B., and Scanlon, Paul W. Ewing's sarcoma; a critical analysis of 165 cases. *J. Bone & Joint Surg.*, Mar., 1961, *43-A*, 185–192. (From: The Section of Surgical Pathology, the Section of Orthopaedic Surgery, and the Section of Therapeutic Radiology, Mayo Clinic and Mayo Foundation, Rochester, Minn.)

Ewing's sarcoma is a highly malignant primary tumor of bone occurring chiefly in the second decade of life and affecting almost any bone. Because the nature of the parent cell is still in marked dispute, the eponym "Ewing's Sarcoma" must still remain to distinguish this specific entity.

For many years after Ewing's original description, the pathologic diagnosis of the condition was indefinite and a variety of neoplasms and some non-neoplastic conditions were included under this diagnosis. With recent advances in the knowledge of bone pathology, the diagnosis is now made with greater exactitude. Ewing's sarcoma may be defined as a highly anaplastic, malignant, small round cell tumor which is primary in bone and usually can be readily differentiated from other small round cell tumors. Occasionally, highly malignant tumors are observed in which the size and morphologic characteristics of the nuclei are intermediate between those of Ewing's sarcoma and those of reticulum cell sarcoma. In such instances, correlation with the clinical findings serves to determine the correct classification. The small cells of Ewing's sarcoma characteristically occur in large islands which contain little or no connective tissue. Grossly, the tumor cannot be definitively distinguished from other tumor or, at times, from inflammatory processes.

The authors report a study of 165 patients with Ewing's sarcoma seen at the Mayo Clinic from 1905 to January 1, 1960, a group comprising somewhat less than 10 per cent of the total number of primary malignant bone tumors seen during the same period. In location, the femur was the most common site, being involved in slightly more than one fourth of the group; approximately three fourths of the tumors occurred in the pelvic girdle and long tubular bones of the extremities. Contrary to the widely-held tenet that Ewing's sarcoma characteristically involves the diaphysis of long bones, it was found that the metaphyseal region was more commonly the epicenter of the growth. In age, the youngest patient was eighteen months and the oldest fifty-nine years. Nearly half of the cases occurred in the second decade and almost 90 per cent within the first three decades. Males predominated by a ratio of 101:64.

Whereas in the past the roentgenographic appearance was considered to be relatively pathognomonic, characterized by osteolysis without osteosclerosis and by an onion-skin layering of the subperiosteal new bone, it was determined in this review that the findings were extremely variable and not specific. Many of the lesions contained zones of sclerosis. Radiating spicules from the cortex of the affected bone were not uncommon. The appearance of Ewing's sarcoma could be mimicked by several other malignant tumors, as well as by eosinophilic granuloma and osteomyelitis. The definitive diagnosis, therefore, had to depend on microscopic examination.

Treatment consisted of irradiation, amputation, local excision, curettage, and Coley's toxins in a wide variety of combinations and for this reason it was difficult to evaluate the effectiveness of any one specific form of treatment. Of 79 patients with

sarcomata in the extremities treated before 1955, 48 received irradiation as the first definitive therapy and 18 underwent amputation as the first measure. The over-all five year survival was 15 per cent and the ten year survival was 10 per cent; 45 per cent of the patients died within the first year and 70 per cent were dead by the end of the second year.

No definite conclusion could be drawn regarding the relative effectiveness of irradiation and ablative surgery. Irradiation was of great value as a palliative measure, particularly for pulmonary metastases, 2 patients surviving more than twenty years after such therapy.—*Leslie K. Sycamore, M.D.*

Ralston, E. L. (Philadelphia, Pa.) Legg-Calvé-Perthes disease—factors in healing. *J. Bone & Joint Surg.*, Mar., 1961, *43-A*, 249–260.

The author reports an analysis of 43 children with unilateral Legg-Calvé-Perthes' disease recognized within three months of onset and treated by a standard method throughout the course. The date of onset is assumed to be the date of the first symptom; the end point is defined as that time at which there is replacement by new bone in the capital epiphysis to the extent that a complete well-defined subchondral layer of bone is visible on both anteroposterior and lateral roentgenograms, and any defect remaining in the epiphysis does not constitute more than 25 per cent of the epiphysis. It is recognized that some degree of remodleing of the head and neck continues up to the time of epiphyseal closure, but the definition permits a precise and standardized period for comparative study.

Chronologic age of the patients varied from four to eleven and one half years with most of the cases falling between four and seven years. Retardation of skeletal age averaged one year and nine months.

Percentage of involvement of the epiphysis, with the author's method of measuring on the roentgenograms, varied from 10 to 100 per cent, with the greatest incidence falling between 50 and 80 per cent. Only 1 case showed complete necrosis of the epiphysis. The duration of the disease varied from eight to fifty-nine months, a wider range than reported by most authors. The largest number of cases fell between twenty and thirty-five months. The degree of recovery, as measured by the comprehensive quotient of Heyman and Herndon, varied from 65 to 100 per cent, with the largest number falling in the 80 to 85 per cent group.

Study of the correlation of the various factors brought out several conclusions somewhat at variance with generally accepted principles. Whereas it has been frequently asserted that the younger the child the more quickly will the disease run its course, in this series there was only a weak correlation. Controverting the theory that the end result is better in the younger child, no such correlation was present between these two factors in this series. A weak correlation existed between the age at onset and the degree of involvement of the epiphysis, the greatest percentage of involvement occurring in the older patients. A high degree of correlation was shown between the extent of involvement of the epiphysis and the duration of the disease, but only a weak correlation was shown between the involvement and the final comprehensive recovery quotient.

The author concludes with a plea for judging the results of treatment on a careful analysis of the roentgen findings rather than on clinical impressions. —*Leslie K. Sycamore, M.D.*

Ponseti, Ignacio V., and Cotton, Ralph L. Legg-Calvé-Perthes disease—pathogenesis and evolution; failure of treatment with l-triiodothyronine. *J. Bone & Joint Surg.*, Mar., 1961, *43-A*, 261–274. (From: The Department of Orthopaedic Surgery, State University of Iowa, Iowa City, Ia.)

The authors observed that the epiphyseal plate lesion in Legg-Calvé-Parthes' disease was similar to the lesion observed in rats and rabbits fed diets containing aminonitriles. Since low doses of l-triiodothyronine suppressed the mild forms of aminoacetonitrile poisoning in rats, it was decided to test the effect of this compound on children with Legg-Calvé-Perthes' disease. Forty-four children of the same age and sex and with similar involvement of the hip were paired, one member of each pair receiving treatment with l-triiodothyronine. Similar orthopedic treatment was used in all cases. The period of treatment with the drug lasted from six to eighteen months.

Biopsy specimens revealed that the bone of the femoral head was necrotic and the epiphyseal plate cartilage was fibrillated and cracked. It was surmised that loss of the blood supply to the femoral head results from the interruption of the retinacular vessels when they cross the cartilage at the edge of the disrupted epiphyseal plate. With revascularization, the dead bone is replaced by living bone, either by the process of creeping substitution or by absorption of the necrotic bone by an advancing front of connective tissue, both processes often being intermingled. Direct creeping substitution is prevalent in the necrotic areas that are structurally well preserved, whereas abundant fibrous tissue is prominently observed adjacent to collapsed necrotic bone. On roentgenograms, structurally well-preserved dead bone is characterized by only slightly increased density. This density increases during the process of replacement by creeping substitution as new bone is laid down on the dead bone trabeculae. Collapsed areas appear on the roentgenogram as blotchy areas of increased density intermingled with more radiolucent areas.

In the earliest cases, the center of ossification of the femoral head is slightly denser and smaller than

on the normal side. Some collapse is present in all patients. An increase in the width of the space between the ossified portion of the femoral head and the inner wall of the acetabulum is observed in most early cases. Collapse occurs in the weight-bearing portion of the femoral head, the posterior portion being spared.

Reconstruction of the necrotic femoral head occurs according to three patterns. Group I (most common type): the posterior third of the epiphysis is rebuilt at a fast rate; the bone density is increased at first and gradually becomes normal in one or two years with new bone being laid down over the dead bone and then slowly replacing it over a period of one or two years; the anterior half ossifies much more slowly than the posterior half with several months elapsing between the absorption of the dense necrotic bone and the formation of radiolucent new bone; the epiphyseal plate is slightly wide and irregular. Group II: the pattern of reconstruction is initially similar to that in Group I; the collapse of the anterior two-thirds of the head, however, is very marked and, after absorption of the dead bone, this portion remains largely unossified for many months and even years. Group III: the bone of the entire femoral head collapses and reossification is very slow and irregular; small areas of new bone reconstruction appear at different sites after several months; the epiphyseal plate is wide and irregular, with large areas of decreased density extending deep into the metaphysis.

The final shape of the femoral head is normal in Group I, slightly flattened in Group II, and extremely flat in Group III. Determination of the degree of severity of the epiphyseal plate lesion is of great prognostic value, the speed of reconstruction being proportional to the severity of this lesion. The width of the femoral neck is increased in all cases, presumably owing to the increase in periosteal new bone formation along the neck. The primary lesion in Legg-Calvé-Perthes' disease appears to be a weakening and disruption of the upper femoral epiphyseal plate. Aseptic necrosis of the femoral head results from the interruption of the retinacular vessels when they cross the cartilage at the edge of the disrupted plate.

Studies of protein bound iodine and of the uptake of active iodine reveal no evidence of thyroid disease and recovery is not altered by the administration of large doses of l-triiodothyronine during periods of from six to eighteen months.—*Leslie K. Sycamore, M.D.*

Brown, B. St.J. The radiologic features of bone changes in tuberous sclerosis with a case report. *J. Canad. A. Radiologists*, Mar., 1961, *12*, 1–9. (From: The Department of Radiology, The Montreal General Hospital, Montreal, Quebec, Canada.)

The author describes the roentgen features of tuberous sclerosis with particular reference to the bone changes. The combination of bone changes in the presence of brain lesions is virtually diagnostic of the condition. In the investigation of epilepsy, mental deficiency and abortive forms of tuberous sclerosis, roentgenograms of the extremities and skull may be of considerable diagnostic value.

Tuberous sclerosis occurs in the general population in about 1 per 150,000, and in institutions for epileptics and mentally defectives in about 0.6 per cent. Tuberous sclerosis is a defect in the histogenesis and organization of tissues with a tendency to the formation of hamartomas. In the literature reviewed by the author there were only four descriptions of pathologic bone lesions in definite cases of tuberous sclerosis.

The roentgen findings occur more frequently in the brain (calcification in 80 per cent), the kidney (tumor-like nodules in 80 per cent), and the hands and feet (cyst-like changes, subperiosteal new bone and cortical pitting in 66 per cent). Patchy areas of increased bone density occurred in the upper halves of the parietal bones in 48 per cent. Less frequently noted were patchy areas of increased bone density in the pelvis and spine. Sclerotic patches and, more rarely, fibrocystic changes were noted in the long bones and ribs.

The differential diagnosis between tuberous sclerosis and osteosclerotic metastases is dependent upon the presence of other types of associated bone changes in tuberous sclerosis. Brain calcification aids considerably in the differential diagnosis.—*J. N. Ané, M.D.*

General

Bistolfi, F., and Pignataro, E. Rapporti fra spessore di strato e nitidezza d'immagini in stratigrafia pluridirezionale. (Relation between thickness of the layer and clearness of the images in multidirectional stratigraphy.) *Radiol. med.*, Dec., 1960, *46*, 1127–1149. (From: Istituto di Radiologia dell'Università di Genova, Italy.)

The recurring problem of the choice of layer thickness in laminagraphy was demonstrated at the last International Congress of Radiology, held in Munich in 1959, where Frain and Gaucher spoke in favor of thin layers and Westra and Ziedses des Plantes in favor of thick layers.

This paper deals with the effect of various layer thicknesses on the clearness of the images in multidirectional laminagraphy. It is based on practical experiments conducted for the past two years in the Institute of Radiology of the University of Genoa, under the guidance of Prof. A. Vallebona.

The technical characteristics of multidirectional laminagraphy can be summarized as follows: (1) shading-off of a material point in a chosen surface; (2) lack of transport shadows; (3) a more complete

shading-off of images than that obtained in unidirectional laminagraphy, resulting in a greater elimination of shadows which are in other layers; (4) longer exposure time (the minimum, in the Pluristrator Zuder used by the authors, is 1.8 sec.); and (5) accurate portrayal of the images in the chosen layer.

The authors have adopted multidirectional laminagraphy in the evaluation of certain parts of the skeletal system, such as articular surfaces, canals and foramina, formations and ossicles of the inner and middle ear, the interruptions along the walls of certain bones (as in the petrous pyramids), mastoid cells (small cells) and paranasal sinuses (large cells), mucosa of the sinuses, and the spongiosa of long and short bones.

From these studies of the skeleton the authors conclude that a thick layer of 5 to 8 mm. gives a clearer image of the structures on the laminagram. The only exception to this general rule is noted on the laminagrams of the spongiosa of the vertebral bodies.—*A. F. Govoni, M.D.*

Campbell, John A., Klatte, Eugene C., Gray, Donald D., and McCrea, Alice L. Evaluation of film size in cineradiography. *Radiology*, April., 1961, *76*, 606–613. (Address: J. A. Campbell, Indiana University Medical Center, 1100 West Michigan Street, Indianapolis 7, Ind.)

Limited image detail remains the most critical obstacle to be overcome if cineroentgenography is to have a wider range of diagnostic usefulness.

The authors evaluate the relative merits of film size alone as a determining factor in the quality of a cine-image. In order to do this, other factors must be controlled: the resolving power of the image tube, image-tube focus, scintillation effect, relative speed of the lens system and focal spot of the roentgen-ray tube.

Image quality is judged by its resolution and its sharpness of detail. The greatest spatial frequency or the narrowest bars and spaces that can be resolved by the use of test objects is said to be the resolving power limit. The various test objects which may be used are the wire screen, General Electric lead reticle, Ter-Pergossian contrast test object, Phillips electric brass reticle, and the Indiana University test pattern used by the authors. This test pattern consists of diverging and converging bars and spaces of varying lines per inch. This arrangement allows testing of the edges of the field as well as the center.

Resolution tests were carried out using various popular 16 and 35 mm. film emulsions with this test pattern and, for comparison, with wire screen cloth and the General Electric lead bar reticle.

The authors conclude that, in clinical cineroentgenography using currently available image intensifiers, medium and fine grain 16 mm. film emulsions can be expected to produce images of diagnostic quality equal to those obtained with fast, coarse grain 35 mm. emulsions with the same radiation dose to the patient.—*W. M. McBride, M.D.*

Boucek, Robert J., Murphy, William P., Jr., and Hernandez, Francisco A. Intercalative angiography; cardiovascular spot-filming. *Radiology*, Apr., 1961, *76*, 565–571. (Address: R. J. Boucek, 1550 N. W. Tenth Avenue, Miami 36, Fla.)

Intercalative angiography or cardiovascular spot-filming is the electronically controlled spot-filming of an opacified portion of the cardiovascular system through the use of electronic devices which are activated by the electrocardiogram to trigger a rapidly acting injector and to trip off the roentgen ray exposure in a predetermined time sequence. Because the volume of the injected radiopaque material is small, high speed injections must be made via a catheter introduced into the area to be visualized so that the bolus of material will be imposed between or within a cardiac cycle, thus producing a discrete localization of the substance at the selected site. This obviates most of the limitations of the standard cardiovascular roentgen-ray procedures and eliminates unnecessary irradiation. In addition, repeat injections may be made during one examination.

A more detailed discussion of the apparatus and the technique, including a few illustrative roentgenograms, is presented.—*Walter H. Jarvis, Jr., M.D.*

Uhr, Nathaniel, and Bezahler, Harvey B. Pseudo-pseudohypoparathyroidism: report of three cases in one family. *Ann. Int. Med.*, Mar., 1961, *54*, 443–451. (From: The C. F. Menninger Memorial Hospital, Topeka, Kan.)

Pseudohypoparathyroidism (PH) was a term used in 1942 by Albright *et al.* to describe a group of patients with round face, short stature, short metacarpals and metatarsals, ectopic ossifications, high serum phosphorus and low serum calcium. This disease is distinguished from idiopathic hypoparathyroidism by failure of the patients to respond to parenteral parathyroid extract with an increased urinary excretion of phosphate. Ten years later Albright described a case with many of the clinical features of PH, but with normal values for serum phosphorus and calcium. He termed this pseudo-pseudohypoparathyroidism (PPH).

The familial tendency of this disease has been noted and the authors report 3 patients, a mother and two sons, meeting most of the clinical and chemical criteria of PPH. Six other cases, first and second cousins of the patients, appeared to bear many superficial resemblances to the reported cases.

The interesting question of the terminology for these diseases is discussed as well as the inter-rela-

tionship between PPH and PH and the probable mode of inheritance.—*George L. Sackett, Jr., M.D.*

RADIOISOTOPES

Sterling, Kenneth, and Tabachnick, Milton. Resin uptake of I^{131}-triiodothyronine as a test of thyroid function. *J. Clin. Endocrinol. & Metabol.*, Apr., 1961, *21*, 456–464. (From: The Department of Internal Medicine, New York State Psychiatric Institute, and the Department of Psychiatry, Columbia University, College of Physicians and Surgeons, New York, N. Y.)

Since Hamolsky and Freedberg and their associates first announced the I^{131} triiodothyronine red blood cell uptake as a measure of the amount of circulating thyroid hormone, many groups have attempted to simplify the test by exposing the patient's serum to an adsorptive resin rather than erythrocytes.

The test described by the authors, while not a simple one to perform, seems the best resin test yet suggested. For those interested in performing it this interesting paper should be read in its entirety, but briefly the method is as follows. The resin IRA-400, one of the amberlite family of resins, is converted from the chloride form, in which it is supplied, to the formate form by washing with 1 M sodium formate solution adjusted to pH 5.0 ± 0.2. The resin is washed until free of chloride ion and stored at 5° C. under sodium formate solution. The resin appears to be stable for at least four months in this form. All operations until the final washings and count are carried out at a temperature of 2–6° C. Two ml. of test serum is added to 0.2 ml. of a solution containing an amount of I^{131} l-triiodothyronine with no more than 0.2 μg. per ml., and with a specific activity appropriate to the counting apparatus used. After shaking in a cold bath for 15 to 45 minutes, 1 ml. aliquots of the serum-triiodothyronine solution are added to tubes containing 1 ml. of washed resin and shaken for 90 minutes at the low temperature. Only during counting are the samples not maintained at a cold temperature. It is pointed out that warming to room temperature is to be avoided, since this increases the resin uptake. In the authors' laboratory, duplicate determinations on a given serum usually agree within 1 to 2 per cent as do the values for a given serum run at different times.

The authors tested the procedure in 35 normal patients and 44 patients with active thyrotoxicosis. Twenty-eight typical cases of myxedema and hypothyroidism were also tested, and values were determined for 11 pregnant women.

Results in the uptake of I^{131} triiodothyronine were: in hypothyroidism, 27 ± 3.3 per cent; euthyroid subjects 35 ± 2.2 per cent; thyrotoxicosis 53 ± 7.8 per cent. In pregnancy these values were depressed to the hypothyroid level or lower. Differences were found to be statistically significant, and apparently more reliable than duplicate determinations of erythrocyte uptake, and then the results by the resin test described by Mitchell. In cases of low TBG, as in the nephrotic syndrome, there is an elevated resin uptake even though the concentration of free thyroxine is normal and the patient is euthyroid. As with other tests of this family results are not influenced by contamination with organic or inorganic iodine.—*Frederick J. Bonte, M.D.*

Bowers, C. Y., Murison, P. J., Gordon, Douglas L., and Locke, William. Effect of thyrotropin on the serum protein-bound iodine level in various thyroid states (TSH-PBI test). *J. Clin. Endocrinol. & Metabol.*, Apr., 1961, *21*, 465–471. (From: The Departments of Medicine, Louisiana State University School of Medicine, Ochsner Clinic, and Tulane University School of Medicine, New Orleans, La.)

Single intramuscular injections of thyroid stimulating hormone (TSH) were administered to 22 normal subject and 67 patients with thyroid disease, including secondary myxedema. Resulting changes in the serum protein-bound iodine (PBI) concentration were measured. Twenty-four hours after injection of 5, 10 or 20 units of TSH the mean rises in PBI concentration for normal subjects were 1.3 μg. ± 0.6 to 2.8 μg. ± 1.0 per ml. In general, rises were abnormally low in patients with diffuse thyroid disease, whereas they were normal in patients with focal thyroid disease or pituitary failure. In some of the patients who had nontoxic nodular goiter, responses were abnormally high. The significance of this phenomenon is not understood.

Because of the fact that TSH raises the PBI and thyroidal I^{131} uptake so little in patients with thyroiditis, the authors suggest the use of a TSH-I^{131} or TSH-PBI test to differentiate Hashimoto's disease from other nontoxic nodular goiters.—*Frederick J. Bonte, M.D.*

Deller, D. J., Germar, H., and Witts, L. J. Effect of food on absorption of radioactive vitamin B_{12}. *Lancet*, Mar. 18, 1961, *1*, 574–577. (From: The Nuffield Department of Clinical Medicine, Radcliffe Infirmary, Oxford, England.)

Baseline determinations were made by administering Co^{58} labelled vitamin B_{12} to 5 normal volunteers, 5 hospital patients with disease not associated with absorption of vitamin B_{12}, 5 patients with pernicious anemia and remission, and 21 patients who had had a partial gastrectomy for peptic ulcer one to eighteen years before the test. These tests were performed in the fasting state, as usual.

The tests were repeated in all of these individuals by giving a test dose of Co^{58} vitamin B_{12} with a vitamin B_{12} free meal.

In the 10 normal subjects and in the 5 who had pernicious anemia the administration of food had no effect, but in the group of patients who had undergone partial gastrectomy the absorption of Co^{58} vitamin B_{12} was significantly enhanced when the test dose was given with food. In some patients in whom the absorption of vitamin B_{12} was grossly subnormal the effect of food was comparable with that of the intrinsic factor. The authors, therefore, conclude that the administration of a fasting dose of radioactive vitamin B_{12} is not a reliable method for determining the ability of the patient to absorb vitamin B_{12} when gastrectomy has been performed.—*Frederick J. Bonte, M.D.*

Duggan, H. E., and Weijer, Dorothy L. Results of treatment of malignant neoplastic disease with radioactive colloidal gold(Au^{198}); an analysis of 60 cases. *Canad. M.A.J.*, May 6, 1961, *84*, 1000–1003. (From: The University Hospital, University of Alberta, Edmonton, Alberta, Canada.)

Radioactive colloidal gold was first used intraperitoneally in 1947 by Hahn. Since then many reports of various workers have appeared in the literature. The method is especially suited for body cavity irradiation. The particle size, .003 to .007 μ in diameter, and the short half life make even irradiation possible with no damage to surrounding tissues. It has been used in the pleural and peritoneal cavity, in cystic brain lesions, and in the prostate.

This paper gives the results in 60 cases treated. The methods used are discussed and graphs showing the irradiation pattern are included. Good results occurred when the gold was used prophylactically at operation for carcinoma of the ovary with no demonstrable implants. However, these results are really not definitive, since the patients might have survived without this treatment.

The over-all results indicate that palliation was the main effect. It had no value in cases of carcinoma of the bronchus.

Sixty patients were treated by instillation of Au^{198} in an attempt to reduce or stop fluid production, diminish tumor size or prevent metastatic spread. Fifty-four had malignant effusions; 6 were treated prophylactically. There were 7 cases of prostatic carcinoma, and 3 of cystic brain tumors. Carcinoma of the ovary with ascites (25 per cent) showed good results with an average survival of nine months. The ovarian carcinomas without ascites showed good results in most cases (5 survived, 1 died). Carcinoma of the prostate (43 per cent) showed some improvement.

This paper is very informative. The results are about the same as those reported elsewhere—palliation only, but the procedure is one more modality of aid.—*Eva S. Carey, M.D.*

Sarasin, Raymond. Les complications de la télécobalthérapie. (Complications of telecobalt therapy.) *Radiol. Clin.*, Nov., 1960, *29*, 346–356. (From: Service de Radiologie de l'Hôpital Universitaire de Genève et du Centre de Télécobalthérapie, Geneva, Switzerland.)

The complications following treatment by high energy radiation did not differ significantly from those observed in conventional radiotherapy. Higher tissue doses, up to 8,000 r, may be employed in some cases, although usually for tumors such as those of the esophagus and bronchus only 4,000 to 5,000 r tissue dose is used for preoperative irradiation and this is usually given in five to seven weeks.

If a tumor which has extensively infiltrated the interstitial tissue is destroyed, this will leave a large region which needs filling in and may actually lead to complication. In addition, there are individual factors: certain patients seem to undergo necrosis while others treated with the same technique do not have any complications.

Preoperative irradiation is used whenever possible for the following reasons: (1) to diminish the degree of activity of the tumor; (2) to diminish the volume of the tumor, which may be of aid to the surgeon; (3) to utilize the more radiosensitive state of the tumor, which becomes less sensitive postoperatively because of changes in vascularity; and (4) to treat the general condition of the patient during irradiation, which, in turn, better prepares him for surgery. Also, patients can scarcely support the effects of irradiation after such operative procedures as pneumonectomy or extensive esophagectomy.—*Charles M. Nice, Jr., M.D., Ph.D.*

MISCELLANEOUS

Lee, J. A. H. Acute myeloid leukaemia in adolescents. *Brit. M. J.*, Apr. 8, 1961, *1*, 988–992. (From: Social Medicine Research Unit of the Medical Research Council, London Hospital, London, England.)

The death rate from leukemia is high in young children, and then falls to a minimum in early adult life. Previous studies show that, in males, the decline in the leukemia death rate during adolescence is interrupted by a transient rise. This excess mortality is not a new phenomenon, but has been observed at least as far back as 1911–15. A study of this phenomenon is now reported.

Analyses of the leukemia mortality data for England and Wales, Scotland, the United States, and for Canada were made. In each of these four countries the decline of the death rate stops at about the

age of 13 in males, and rises to a new peak at about 17. A further decline then occurs and continues until the late twenties. In females there is evidence of a similar trend, but it is less marked. In the United States and Canada the rise in the mortality rate in females appears to occur earlier than in males.

The data from England and Wales suggest that the death rate from leukemia in males aged 15–19 is about 6 per million per annum greater than would be expected if the trend of the rates with age were not interrupted in this period, The male population of this country in this age group is 1.4 million, so that the whole effect described in this paper is due to about 10 cases occurring annually. This figure gives a measure of the difficulty of studying the problem. However, the smallness of the numbers is due to the rarity of leukemia of any sort rather than to the smallness of the rate increase in adolescents. There is no evidence on which to base any discussion of the relationship of these cases of leukemia to radiation.

The high leukemia rates in male adolescents are matched by corresponding increases in admissions to hospitals for acute infections of the lower respiratory tract. In females the rise in the number of hospital admissions for these conditions is similar, but, as already pointed out, in England and Wales at least, the change in the female leukemia rate is less marked. These are interesting findings, because in children under the age of 10, Stewart (1961) has put forward evidence that pneumonia may induce leukemia after a short latent period.—*George Tievsky, M.D.*

Court Brown, W. M., and Doll, R. Leukaemia in childhood and young adult life; trends in mortality in relation to aetiology. *Brit. M. J.*, Apr. 8, 1961, *1*, 981–988. (Address: W. M. Court Brown, Director of the Medical Research Council's Clinical Effects of Radiation Research Unit, Western General Hospital, Edinburgh, Scotland.)

There is good reason to believe that the different clinical and cytologic types of leukemia represent distinct diseases. Thus, there are major differences between the age and sex distributions of chronic lymphatic, chronic myeloid, and acute leukemia. Furthermore, ionizing radiations have been shown to cause chronic myeloid and acute leukemia, but they have not been shown to cause chronic lymphatic leukemia, and the geographical distribution of the first two types differs from that of the last in that chronic lymphatic leukemia is relatively rare in the Far East. Finally, recent cytogenetic data have demonstrated the existence of a specific chromosomal abnormality in many cases of chronic myeloid leukemia, but, to date, not in any other type.

An analysis has been made of the mortality rates from leukemia among children and young adults under the age of 30 years in England and Wales during the period 1911–1959. Similar data for other countries have been examined over a shorter period.

Estimated mortality rates for acute lymphatic leukemia showed a maximum at the age of 3 years in both sexes. The mortality rate fell to a minimum at ages 25–29 years, with only a slight check at ages 15–19 years. The mortality from acute myeloid leukemia varied less sharply, but showed two peaks —at ages 2–4 years and 15–19 years. The reduction in the mortality figures for acute lymphatic leukemia in late adolescence and the peak in the childhood mortality rate from acute myeloid leukemia may be due to misclassification. The mortality from both types of chronic leukemia was low throughout the age range studied, but there was evidence of an increase in the mortality from chronic myeloid leukemia above the age of 20 years.

Between 1911 and 1959 the mortality increased fairly steadily in each five year age group and in each sex. The average rate of increase was approximately 4.5 per cent per annum under the age of 10 years and slightly less than 4 per cent per annum at ages 20–29 years. The age groups 0–4 years and 15–19 years showed no increase in the last 10 years. Part of the increase is likely to be spurious and due to such factors as better case-finding and improvements in the therapy of infections; but it is difficult to account for the continued increase at young adult ages by these factors alone. Data for the last 15 years suggest that there has been a real increase in chronic myeloid leukemia and in acute myeloid leukemia over the age of 15 years.

Before 1920 there was no evidence of a childhood peak in the mortality from leukemia. A peak mortality was present at ages 2–3 years from 1921 to 1939, and since 1940 the peak has become more pronounced. Mortality rates for the white population of the U.S.A. since 1940 have been similar, but at a higher level. Peaks in the childhood mortality have not been observed in the nonwhite population of the U.S.A. or in Japan.—*George Tievsky, M.D.*

Garland, L. Henry. Cancer of the thyroid and previous irradiation. *Surg., Gynec. & Obst.*, May, 1961, *112*, 564–566. (From: The Department of Radiology, University of California, Medical School and San Francisco General Hospital, San Francisco, Calif.)

The author compares statistics of several investigators and finds that the claim that previous irradiation about the nasopharynx and mediastinum contributes to development of carcinoma of the thyroid has no real proof. These comparative studies are impressive in their results.

Clark in Chicago encountered 13 cases of thyroid carcinoma in children who had a history of irradiation. The calculated dose was about 18–60 r. Uhlmann followed 480 of 2,500 children who had had previous irradiation and he found no carcinoma. Simpson and associates reported 1 case in an infant

to whom 3,000 r was delivered. This dose was obviously excessive and is conceivably related to the subsequent neoplasm. Snegireff was unable to find any cases of leukemia or thyroid cancer in cases with a history of previous irradiation. Latourette and Hodges studied 867 patients who had had thymic irradiation and they found 1 case of thyroid cancer contrasted to a normal expectancy of 0.06. Conti and Patton studied 1,564 irradiated cases. Evidence of subsequent carcinoma was thoroughly searched for and, even though 4 cases would normally be expected, no case was found; 1 case of leukemia was expected, but none was found.

The author estimates that during the years 1925 to 1945 at least 4,000,000 infants received irradiation to the mediastinum, either for treatment or during examination. The death rate for carcinoma of the thyroid up to 1958 has been static.

From a search of all available literature one must conclude that an etiologic connection between low dose neck irradiation and subsequent thyroid cancer cannot be regarded as established.—*Eva S. Carey, M.D.*

Garibaldi, G., and Gugliantini, P. Il problema del trattamento della iperplasia timica in rapporto ai supposti pericoli della roentgenterapia e ai risultati della cura medica. (The problem of treatment of thymic hyperplasia in relation to the supposed dangers of roentgen therapy and to the results from medical therapy.) *Radiol. med.*, Dec., 1960, *46*, 1159–1182. (From: Reparto di Radiologia Ospedale Pediatrico del Bambino Gesù, Rome, Italy.)

In the first half of this century the treatment for thymic hyperplasia was in the majority of cases limited to roentgen therapy; the first report dates back to a paper published by Friedlander in 1907 (Status Lymphaticus and Enlargement of the thymus with a Report of a Case Successfully Treated by the X-ray. *Arch. Pediatr.*, 1907, *24*, 490–501). In analyzing the several statistics on this matter the authors note that, at least in the United States, the highest peak in the use of this treatment was reached in the years 1932–1941 and that the number of cases so treated slowly diminished until, in 1950, roentgen therapy for thymic enlargement was almost completely abandoned. A similar attitude, although to a lesser extent, was noted in Europe.

The principal arguments against the use of ionizing radiation in the treatment of thymic enlargement are the following: (1) through the years radiologists have become aware of the fact that in a number of cases an apparent widening of the superior mediastinum is not secondary to an enlarged thymus but to artefacts related to the position of the patient, stage of inspiration, etc.; (2) not all thymic hyperplasias are accompanied by symptoms and, therefore, there is no need to treat such cases; (3) the usefulness of the adrenocorticosteroids in the treatment of this condition has been demonstrated; and (4) several papers have been published on the late appearance of carcinoma, particularly of the thyroid gland, in individuals previously treated by roentgen rays for thymic enlargement.

The authors review and discuss this topic as it is evaluated today, particularly in the light of the most recent information published in the American literature.

In relation to the roentgen diagnosis of thymic hyperplasia, the authors analyze the technical factors which not infrequently lead to the incorrect diagnosis of enlargement of the thymus, and the other pathologic conditions of the thymus which could be incorrectly interpreted as hyperplasia.

It is well known that in about 60 per cent of the cases an enlargement of the thymus does not produce any clinical symptoms. In these cases and in a few others in which symptomatology is minimal, there is no need for medical or roentgen therapy. The authors point out that the clinical picture, and not the roentgen findings, should determine whether to consider any treatment.

Several papers in recent medical literature deal with the use of adrenocorticosteroids. The immediate results obtained by the authors are very satisfactory but after some time the symptoms tend to reappear in a certain number of cases. To obtain a satisfactory result one must administer a dose of 5 mg. of ACTH, or 1.5–2 mg. per kilogram of prednisolone *per diem*. These dosages are not without danger and, in the authors' opinion, adrenocorticosteroid therapy in infants is rather hazardous, particularly, if the treatment is prolonged.

Over the past ten years, and almost exclusively in the American literature, several papers have pointed out the relationship between roentgen therapy for thymic hyperplasia and cancer of the thyroid. It seems to the authors, however, that one reason for this high incidence is the different technique of irradiation adopted in the United States, where rather large fields have been used. In European Departments of Radiotherapy the emphasis was and is on the use of very small fields, so that the thyroid gland cannot be included in the portal of entry of the radiation beam. Latourette and Hodges report a series in which there was only 1 case of thyroid carcinoma, and that in a patient who was irradiated through a large field. Similar conclusions on the importance of a small field were drawn by Conti *et al.* in 1960.

The authors conclude by recommending roentgen therapy for only those cases in which it is indicated after a complete evaluation of the clinical and roentgen findings. The importance of small fields is emphasized.—*A. F. Govoni, M.D.*

Mathé, G., Amiel, J.-L., and Bernard, Jean. Traitement de souris AkR à l'âge de six mois par irradiation totale suivie de transfusion de cellules hématopoiétiques allogéniques. Incidences respectives de la leucémie et du syndrome secondaire. (Treatment of AkR mice at the age of six months by total irradiation followed by transfusion of allogenic hematopoietic cells. Respective incidence of leukemia and the secondary syndrome.) *Bull Assoc. franç. de l'étude du cancer*, July–Sept., 1960, *47*, 331–340. (From Centre de Recherches de l'Association Claude-Bernard, Hôpital Saint-Louis, Paris, France.)

In this experiment, mice of the AkR strain showed spontaneous induction of leukemia. These mice were treated at the age of six months by a lethal dose of irradiation followed by a transfusion of myeloid cells of adult or fetal C_3H or $C_{57}Bl_6$. The control mice died of leukemia between the sixth and fourteenth months (median, eight months and eighteen days). The mice treated as above also died between the sixth and fourteenth months but the median age at death was seven months, thirteen days. Apparently these animals did not die of leukemia but, rather, died of the changes induced by the treatment. The secondary syndrome includes lymphoid aplasia, hyperplasia of pyroninophilic cells and necrotic changes in the liver and spleen.—*Charles M. Nice, Jr., M.D.*

Boyd, Elizabeth, Buchanan, W. Watson, and Lennox, Bernard. Damage to chromosomes by therapeutic doses of radioiodine. *Lancet*, 1961, *1*, 977–978. (From: Western Infirmary, Glasgow, Scotland.)

Last October, Tough, Buckton, Baikie and Court Brown reported that roentgen rays applied to the spine in the relatively heavy doses used in treatment of spondylitis ankylopoietica produced a shower of chromosomal abnormalities in cultures of the blood. This observation opens up the possibility of a biologic comparison of the effect on the marrow of different forms of irradiation. The authors point out that very little is known of the dose actually received by structures other than the thyroid, when radioiodine is administered in treatment dosage for the thyroid. It was decided to try to find out whether treatment with radioiodine had an effect on chromosomes similar to that of spinal irradiation.

Tough and associates noted a fall in the proportion of cells with the normal chromosome number, and the appearance of chromosomes with structural abnormalities. These structural changes consist of dicentric abnormalities and fragments. These are both extremely rare in nonirradiated cultures. The authors state that they have never seen a dicentric chromosome in any other material. They also believe that there is some indication that structural abnormalities appear later and persist longer than abnormalities in number. They feel that it is already justifiable to conclude that radioiodine can produce changes not very different in degree from those described by Tough and associates as following a single dose of 250 rads to the spine. These findings raise the question whether radioiodine therapy can cause leukemia. In a few reported cases leukemia has followed such treatment. Pochin made a very extensive survey of this treatment in thyrotoxicosis, and concluded that the incidence of leukemia could have been due to chance. However, the larger doses used for thyroid cancer metastases may well carry a more substantial risk, especially when they are repeated several times.

Six brief case reports of patients treated with radioiodine are included, with a chart of chromosome counts.—*Douglas S. Kellogg, M.D.*

Stewart, John S. S., and Sanderson, Ann R. Chromosomal aberration after diagnostic x-irradiation. *Lancet*, 1961, *1*, 978–979. (From: Department of Surgery, and Department of Zoology, Queen's College, Dundee, Scotland.)

Chromosome damage in man induced by therapeutic irradiation has recently been reported by Tough and associates. The authors have observed a similar type of chromosomal aberration in leukocyte culture of the peripheral blood from a patient with medullary dysgenesis (chromatin-positive Klinefelter's syndrome) shortly after exposure to diagnostic irradiation. The report of Tough and associates that dicentrics and other chromosome aberrations may be induced by therapeutic irradiation *in vivo* prompted the authors to recheck their data. It was recalled that the patient had been exposed to diagnostic roentgen rays before the blood sample was taken for culture.

A blood sample was used to set up a culture of leukocytes, which was harvested three days later. Thirty-one plates of high technical quality were suitable for analysis. In 29 the sex chromosome constitution was XXY, and the chromosome number was 47. In the other two plates an abnormally long and apparently dicentric chromosome was present. The aberration might have been induced by the diagnostic irradiation *in vivo* or by background radiation. It is also possible that it might be related to patient's abnormal karyotype, or it could have arisen in culture. Further work is necessary to investigate these possibilities.—*Douglas S. Kellogg, M.D.*